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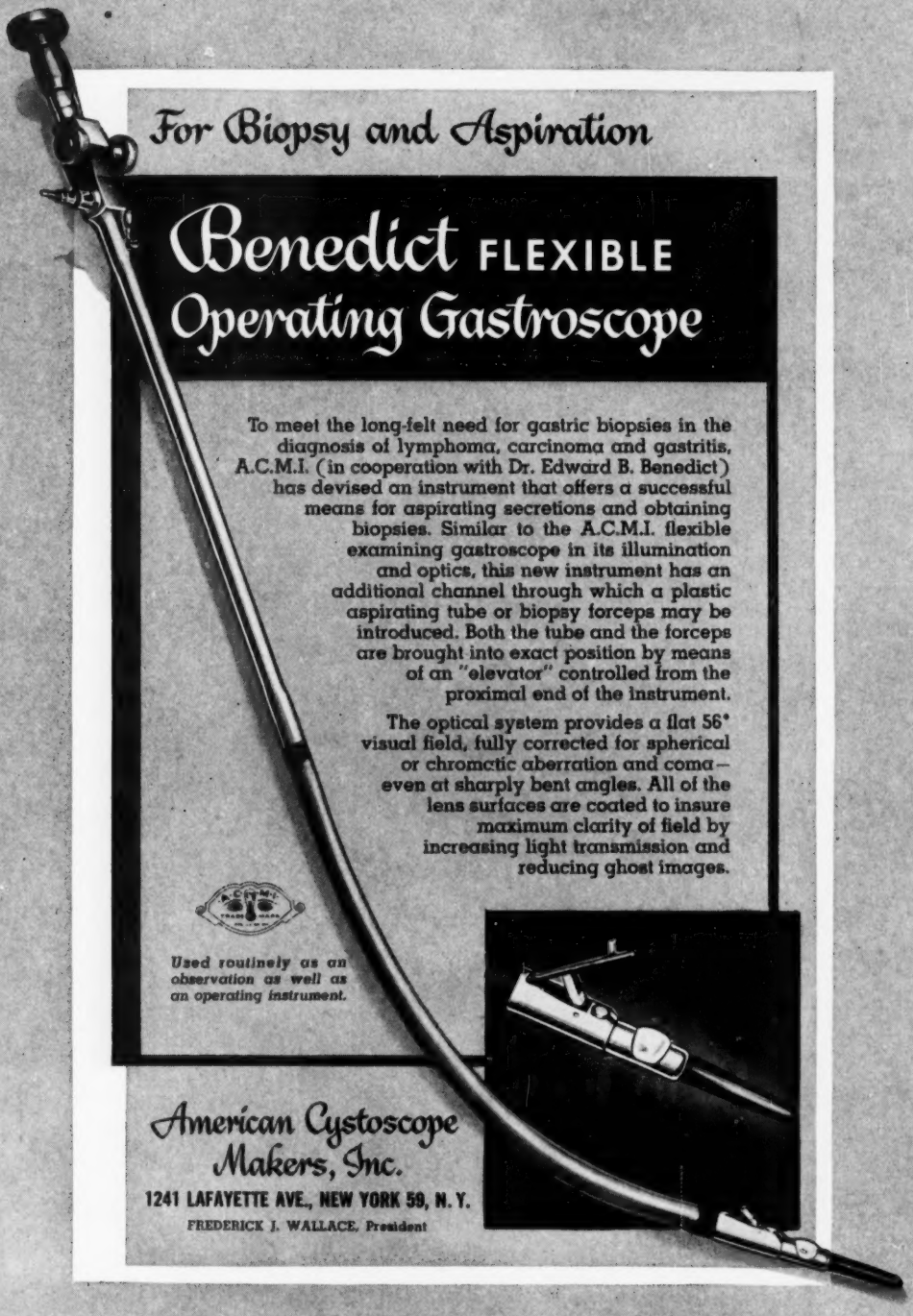

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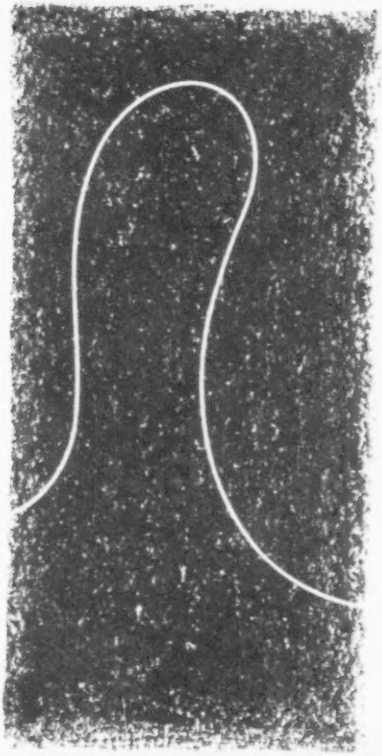

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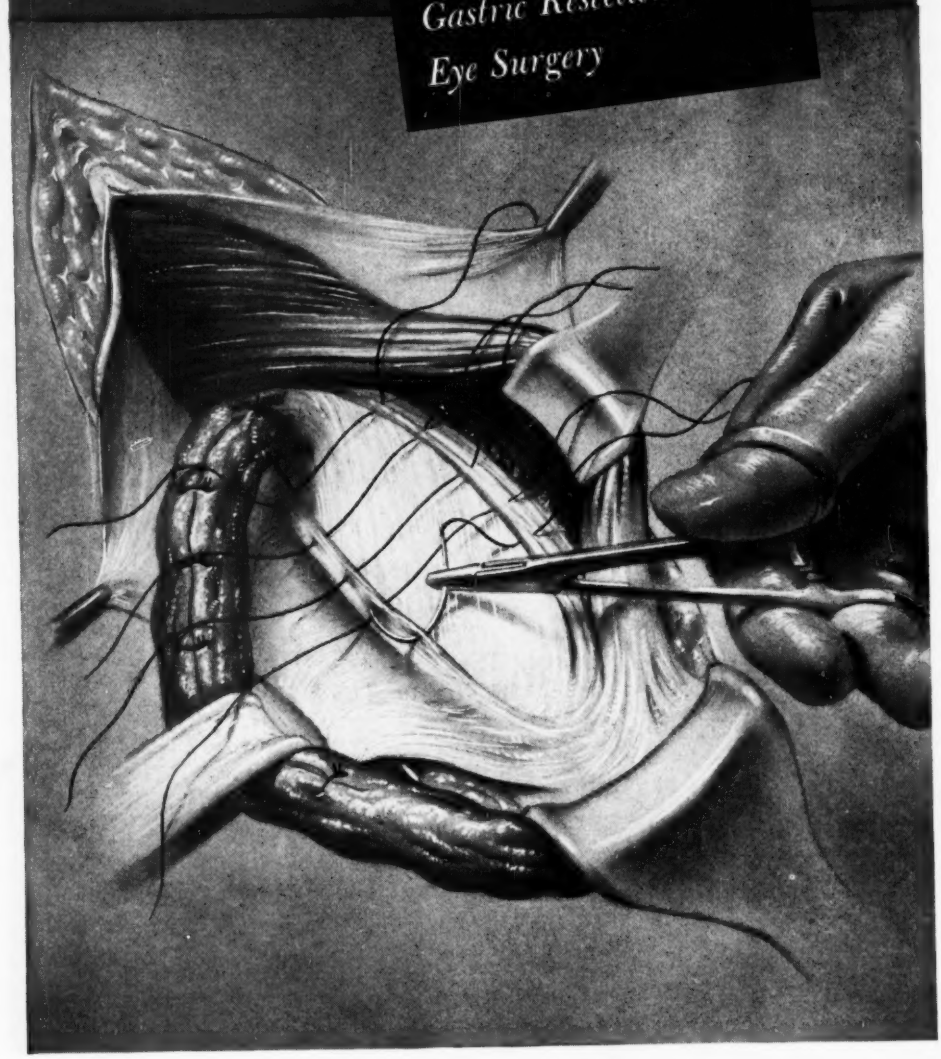

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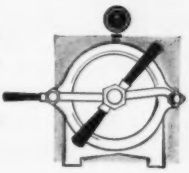

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1. Ferguson, C., and Miller, C. D.: J. Urol. 67:702 (May) 1952.
2. Trafton, H. M., and Lind, H. E.: Ibid. 69:215 (Feb.) 1953.
3. Bisbey, F. B.: Canad. M. A. J. 66:251 (Feb.) 1952.

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ARCHIVES OF SURGERY

VOLUME 67 OCTOBER 1953 NUMBER 4

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Editorials

NEW LIGHT ON THE PHYSIOLOGY OF THE GASTRIC ANTRUM

AS EARLY as 1906, Edkins [1] reported that the intravenous administration of extracts of the mucous membrane of the antrum of the stomach stimulated gastric secretion, whereas similar extracts made from other parts of the stomach had little or no effect. In 1909, Edkins and Tweedy [2] reported that the secretion of acid by the fundus of the stomach could be stimulated by introducing various food substances into the antrum which had been separated from the fundus by a diaphragm. As a result of these studies, and influenced, no doubt, by the recent work of Bayliss and Starling on pancreatic secretin, Edkins postulated that this stimulation of gastric secretion was due to a hormone which he named gastrin, and which was produced by the mucosa of the antrum in response to chemical stimulation by foodstuffs.

This work of Edkins was not widely known and probably played no part in the introduction of partial gastrectomy as a treatment for duodenal ulcer by the surgeons of Vienna and Budapest several years later. Some of these surgeons believed that the beneficial effects of partial gastrectomy were due to the removal of acid-secreting gastric mucosa, although as early as 1912, Harvey and Bensley had demonstrated by staining methods the absence of acid-secreting cells in the mucous membrane of the antrum of the stomach. In the early years, it is probable that the type of gastrectomy employed removed the antrum and only a small fraction of the acid-secreting mucosa of the body of the stomach. Doubt concerning the validity of the gastrin hypothesis of Edkins arose among physiologists with the finding by Ivy and Whitlow,[3] and Priestley and Mann [4] that the application of foodstuffs to isolated antrum pouches in dogs failed to stimulate gastric secretion. These various studies led to the view that whereas the secretion of gastric juice, which resulted from the presence of food in the stomach or intestines of animals after complete vagotomy, was due to stimulation of the gastric glands by a humoral agent, this was believed to arise from the mucous membrane of the stomach and intestine and not specifically from the antrum. On the other hand, surgeons who had noted the high incidence of marginal ulcer following gastrectomy in which the antrum was excluded by the method of Finsterer and Devine, came to regard the antrum mucosa as of special importance.

From the Department of Surgery of The University of Chicago, The School of Medicine.

1. Edkins, J. S.: J. Physiol. **34**:133, 1906.
2. Edkins, J. S., and Tweedy, J. N.: J. Physiol. **38**:263, 1909.
3. Ivy, A. C., and Whitlow, J. E.: Am. J. Physiol. **60**:578, 1922.
4. Priestley, J. T., and Mann, F. C.: Arch. Surg. **25**:395, 1932.

Recent work [5] in our laboratory has provided strong support for the gastrin theory, and has established that the antrum is an endocrine organ, distinct in function from the mucosa of other parts of the gastrointestinal tract, and which under appropriate conditions liberates into the blood stream chemical substances which may either stimulate profoundly the secretion of gastric juice, or inhibit its formation. Excision of the antrum was found to reduce the secretion from vagus denervated gastric pouches in dogs to a negligible quantity. Since in these animals food still comes in contact with the mucous membrane of the esophagus, body of the stomach, and small intestines, it is clear that these tissues do not furnish gastric secretory hormones and that the secretion of gastric juice may be practically abolished by vagotomy and antrum resection without removing any of the acid-secreting mucosa. The application of food substances to isolated antrum pouches was found to stimulate gastric secretion provided the foods were not acid in reaction. Indeed, the application of acid solutions to the antrum mucosa caused an inhibition of gastric secretion from vagus denervated pouches, thus indicating that the effect must be mediated by chemical substances from the antrum which have a depressant effect. Wilhelmj [6] had previously reported that when the acidity of the gastric contents approaches 60 clinical units, inhibitory processes within the stomach are set in action which operate to decrease the rate of secretion of gastric juice due to the chemical phase of secretion.

These findings indicate that the stomach exerts a direct effect on gastric secretion providing juice for the digestion of food and curtailing the secretion when it might become harmful to the mucous membrance. Under normal conditions in the interval between meals, a continuous secretion, small in amount, is regularly found. During the ingestion of food, stimulation of the gastric glands is brought about reflexly over the vagus nerves by the sight, odor, and taste of food and the secretion is continued by the contact of food with the antrum of the stomach. Gastric secretion continues until the gastric contents become acid in reaction, perhaps at a concentration of 60 clinical units of free acid as suggested by Wilhelmj, whereupon, inhibitory chemical substances are released by the antrum mucosa which stop further secretion.

In some of the methods of surgical treatment of peptic ulcer, the function of the antrum is altered or removed altogether. It is thus evident that a better knowledge of the physiology of this endocrine organ under normal and pathological conditions is of great practical as well as theoretical importance.

LESTER R. DRAGSTEDT, M.D.

5. Dragstedt, L. R.; Woodward, E. R.; Storer, E. H.; Oberhelman, H. A., and Smith, C. A.: Ann. Surg. **132**:626, 1950; Am. J. Physiol. **165**:386, 1951. Oberhelman, H. A.; Woodward, E. R.; Zubiran, J. M., and Dragstedt, L. R.: Am. J. Physiol. **169**:738, 1952.

6. Wilhelmj, C. M.; McCarthy, H. H., and Hill, F. C.: Am. J. Physiol. **118**:766, 1937.

HÜRTHLE CELL CARCINOMA

IRA S. GOLDENBERG, M.D.
NEW HAVEN, CONN.

THE SO-CALLED Hürthle cell tumor of the thyroid represents one of the less common neoplasms of this gland. There are relatively few case reports [1] of this malignant tumor as compared with numerous reports of other types of carcinoma of the thyroid. The lesion is of interest from the varied viewpoints of etiology, pathology, and treatment. It is the purpose of this study to report 22 cases of Hürthle cell carcinoma diagnosed and treated on the Yale University Surgical Service and the Grace-New Haven Community Hospital during the years 1927 to 1952 inclusive.

The commonly accepted term "Hürthle cell tumor" is a misnomer, and only prolonged usage with subsequent acceptance has prevented the name from being abandoned. The first reported demonstration of the large cells with finely granular, eosinophilic cytoplasm peculiar to this lesion was in 1881 by Baber.[2] It was not until 1894 that Hürthle [3] demonstrated these cells in the thyroid glands of young dogs in a parafollicular location. Accumulations of these cells with pale, vesicular nuclei forming definite tumefactions were first reported by Langhans in 1907,[4] but it was Ewing [5] who first used the term "Hürthle cell tumor" in referring to encapsulated tumors of this specific cell type.

In the past the lesion was thought to represent a benign tumor which had malignant potential and was classified in this manner by Warren.[6] More recently,

From the Department of Surgery (Oncology), Yale University School of Medicine, and the Grace-New Haven Community Hospital.

1. (*a*) Bakay, L., Jr.: Parafollicular Cell Adenoma of the Thyroid Gland, Arch. Path. **45**:447-453, 1948. (*b*) Chesky, V. E.; Dreese, W. C., and Hellwig, C. A.: Hürthle Cell Tumors of the Thyroid Gland: A Report on 25 Cases, J. Clin. Endocrinol. **11**:1535-1548, 1951. (*c*) Ficarra, B. J.: Hürthle Cell Adenoma of the Thyroid, New York J. Med. **52**:1176, 1952. (*d*) Frazell, E. L., and Duffy, B. J., Jr.: Hürthle-Cell Cancer of the Thyroid: A Review of 40 Cases, Cancer **4**:952-956, 1951. (*e*) Morrow, W. J.: Hürthle Cell Tumor of the Thyroid Gland in an Infant, Arch. Path. **40**:387-391, 1945. (*f*) Neidhardt, H. W.: Two Unusual Lesions of the Thyroid Gland, Texas J. Med. **45**:46-49, 1949. (*g*) Sheiman, L., and Kravchick, D. I.: Hürthle Cell Tumor, New York J. Med. **50**:1275-1277, 1950.

2. Baber, E. C.: Researches on the Minute Structure of the Thyroid Gland, Phil. Tr. Lond. **172**:577-608, 1881-1882.

3. Hürthle, K.: Beiträge zur Kenntnis des Sekretionsvorganges in der Schilddrüse, Arch. ges. Physiol. **56**:1-44, 1894.

4. Langhans, T.: Über die epithelialen Formen der malignen Struma, Arch. path. Anat. **189**:69-152, 1907.

5. Ewing, J.: Neoplastic Disease: A Treatise on Tumors, Ed. 3, Philadelphia, W. B. Saunders Company, 1928, pp. 952-953.

6. Warren, S.: The Classification of Tumors of the Thyroid, Am. J. Roentgenol. **46**:447-450, 1941.

however, there has been a realignment of thought, and the tumor is now considered to be malignant. The most recent edition of the "Manual of Tumor Nomenclature," prepared by the American Cancer Society in 1951,[7] has abandoned the designations "Hürthle cell tumor" and "Hürthle cell adenoma" as being obsolete and vague, and suggests that the terms no longer be used. Instead, the malignant nature is emphasized and the designations "Hürthle cell adenocarcinoma" and "Hürthle cell carcinoma" are recommended. The term "Hürthle cell" will be used throughout the present report to avoid confusion.

ETIOLOGY

Careful examination of histological preparations of almost any thyroid gland will usually reveal the presence of at least a few large, polyhedral, acidophilic cells of the Hürthle cell type. Wilensky and Kaufman [8] pointed out many years ago the possibility that the appearance of Hürthle cells indicated a physiopathological rather than an anatomical entity. Friedman [9] was of the opinion that thyroid involution was important. He divided involution into two phases—follicular and cellular. In the former the predominant process was redeposition of follicular colloid with return of the glandular epithelium to the flattened resting state after a period of hyperplasia. The latter, in contrast, referred to the formation of eosinophilic cellular elements (Hürthle cells) in thyroid conditions considered to be regressive, such as myxedema, "exhaustion atrophy," exophthalmic goiter, and Hashimoto's disease. The cellular change may be taken as an indication that the new cells represent elements "burned out" as a result of overstimulation, according to Friedman.

Hamperl [10] has postulated that cells which he called "onkocytes" are important. These cells may be easily demonstrated by the inclusion stains of Feyrter. They are said to represent degenerative transformation of parenchymal cells of the thyroid, and are found as well in the parathyroid, adrenal, salivary, and pituitary glands. Onkocytic transformation in the thyroid may occur in adenoma and carcinoma as well as in the normal thyroid. According to Hamperl, there are two other cell types also found in the thyroid—the colloid cells of Langendorff and the interfollicullar (parafollicular) cells of Hürthle. Because of the similarity in these cell types, onkocytic, microfollicular adenomas or carcinomas are incorrectly designated as Hürthle cell tumors. Bakay [10a] described three cases of goiter with adenomas containing a cell type similar to the onkocyte of Hamperl, but he considered the onkocyte to be a degenerated variety of the parafollicular cells of thyroid.

7. Manual of Tumor Nomenclature and Coding, prepared by the Subcommittee of the Statistics Committee, American Cancer Society, New York, 1951.

8. Wilensky, A. O., and Kaufman, P. A.: Hürthle Cell Tumor of the Thyroid Gland, Surg., Gynec. & Obst. **66**:1-10, 1938.

9. Friedman, N. B.: Cellular Involution in the Thyroid Gland: Significance of Hürthle Cells in Myxedema, Exhaustion Atrophy, Hashimoto's Disease, and Reactions to Irradiation, Thiouracil Therapy, and Subtotal Resection, J. Clin. Endocrinol. **9**:874-882, 1949.

10. Hamperl, H.: Onkocytes and the So-Called Hürthle Cell Tumor, Arch. Path. **49**:563-567, 1950.

Other workers have suggested a possible derivation of the Hürthle cell type from remnants of the lateral thyroid anlage, from postbranchial remnants,[11] or from ectopic parathyroid cells.[12]

PRESENTATION OF DATA

Incidence.—Hürthle cell carcinoma occurred in 1.7% of 1,330 surgical specimens of thyroid gland excised during the years 1927 to 1952 inclusive on the University Surgical Service. This resembles the experience of Chesky and others,[1b] who found an over-all incidence of slightly greater than 1% in their surgical thyroid specimens. Solitary adenomas numbered 486 in the present series and malignant thyroid tumors were found in 73 patients. The incidence of Hürthle cell carcinoma was therefore 4.5% of solitary adenomas surgically excised and 30% of the malignant thyroid tumors. Chesky and his associates [1b] reported that approximately 5% of all solitary adenomas in their series were of the Hürthle cell type, while Frazell and Duffy [1d] reported that at Memorial Hospital in New York City 10% of malignant thyroid tumors were of this type.

Age and Sex.—Hürthle cell carcinomas are most commonly found in women over the age of 40 years. In the present series 86% of the patients were female,

TABLE 1.—*Data from Reported Series of Hürthle Cell Carcinoma (Including Present Series)*

	Cases	Age	Sex Female %	Hyperthyroidism Total %	Hyperthyroidism Previous Therapy %	Distant Metastases and/or Recurrence %	Radical Surgery %	Local Surgery %
Chesky [1b]	25	Average 50	96	36	12	12	4	100
Morrow [1e]	23	70% over 40	85	39	20	20	0	78
Frazell and Duffy [1d]	40	Average 50	68	?	42	40	25	38
Present series	22	82% over 40	86	23	22	5	5	100

and 82% were more than 40 years of age. The youngest patient was 16 years and the eldest was 75 years of age (Table 1).

Symmers [13] reported a case of an infant in whom a gross tumor (later proved to be of the Hürthle cell type) was present from birth, and Morrow [1e] added a case of a child 2 months of age with the same type of tumor. Morrow believed that these tumors could best be explained as having arisen from an overgrowth of embryonal thyroid cells.

Clinical Course.—This tumor behaves much the same as a solitary adenoma of the thyroid. It usually occurs in patients who have no antecedent history of thyroid dysfunction. A few patients display clinical hyperthyroidism. In the present series they were five in number. In four of these patients a diagnoses of hyperthyroidism had previously been made, and they had been treated with iodine preparations and/or thiouracil compounds. In addition, two of the patients had undergone subtotal thyroidectomy because of toxic symptoms unrelieved by drugs. Hürthle cell carcinoma was not present in surgical specimens from either of these patients (Table 1).

11. Getzowa, S.: Über die Glandula parathyreoidea intrathyreoideale Zellhaufen derselben und Reste des postbranchialen Körpers, Arch. path. Anat. **188**:181, 1907.

12. Eisenberg, A. A., and Wallerstein, H.: Hürthle Cell Tumor, Arch. Path. **13**:716-724, 1932.

13. Symmers, D.: Congenital Hürthle Cell Tumor, Arch. Path. **31**:99-102, 1941.

A mass in the neck (present from five weeks to 20 years before medical attention was sought) was noted in 77%, and pressure symptoms (dysphagia and/or dyspnea) were the first symptoms of abnormality in 41% of patients in the present series. Four patients were asymptomatic, a thyroid nodule being discovered in two patients on routine physical examination for other reasons; the remaining two patients had substernal thyroid enlargement noted on screening chest roentgenograms.

Physical examination revealed diffuse thyroid enlargement in 60%, while multiple nodular enlargement was noted in 13% of the present series. A solitary thyroid nodule was present in eight patients (contralateral diffuse enlargement was present in two of these patients also). Palpable cervical lymph nodes were noted in two patients, but in only one patient did these ultimately prove to be metastatic disease. Distant metastases were found in no patient during the first visit and in only one patient in the series did they eventually develop.

Laboratory examinations such as determination of serum iodine level and basal metabolic rate proved disappointing, since no definite abnormal trend was noted. In the present group 5 of 7 serum iodine determinations and 8 of 14 basal metabolic rate determinations showed normal values. When chest roentgenograms (with or without esophageal studies) are made, abnormalities are almost always noted. In 13 patients in the present series such films revealed abnormalities (esophageal pressure defects, tracheal shift, substernal mass) in all of them.

Pathology.—There is nothing striking about the gross appearance of these tumors. They closely resemble benign adenomas, usually being encapsulated. The relative malignancy of many tumors can be estimated by microscopic examination of the tumor. Although the ultimate behavior of the tumor is not necessarily correlated with the histology, prognosis can often be estimated. Thyroid tumors, however, show very poor correlation between these factors, and the Hürthle cell carcinoma particularly is unreliable in this respect. Frazell and Duffy [1d] believe that vein invasion is an important factor in determining prognosis, and they state that it is invariably present in those cases in which osseous metastases develop. Foote, cited by Chesky and others,[1b] also believes that vein invasion is important, but includes as equally important such factors as cellular incidence of malignancy (atypical cells, mitoses, etc.). He divides the tumors according to cellular and nuclear characteristics into orderly and disorderly types. The orderly form may metastasize but usually runs a benign course, while the disorderly form is quite unpredictable. Chesky and associates [1b] believe that, in general, Hürthle cell carcinomas behave much like solitary benign adenomas.

The cellular pattern of Hürthle cell carcinomas can be divided into four categories: [1b]

1. Trabecular (solid cords of cells, two to six cells wide, separated by scanty stroma).
2. Alveolar (small glandular spaces).
3. Papillary (stalks of connective tissues covered by a single layer of tumor cells).
4. Large follicular (large glandular spaces often filled with thin colloid).

Chesky and his associates [1b] noted that the majority of the tumors in their series were of a mixed variety, with cellular elements of at least two types present. In

those patients with toxic symptoms the surrounding tissue was usually hyperplastic, and these writers believe that the toxic symptoms have their origin in the hormone produced in these areas rather than by the tumor.

In the present series, nine tumors were of the trabecular type, seven of the alveolar type, and two of the papillary variety; the remaining four tumors were of the mixed category, showing trabecular, alveolar, and large follicular elements in various combinations (Figure). Blood vessel invasion was present in eight tumors, but in only one case were distant metastases noted.

Treatment.—Treatment of this lesion can follow a clear-cut pattern only after a decision has been reached regarding the malignancy of the tumor. In almost no case is Hürthle cell carcinoma diagnosed preoperatively, although carcinoma may be suspected clinically before operation. In some cases frozen section examination with positive diagnosis may be followed by primary radical surgery, but in most cases local excision alone is contemplated and carried out because the true diagnosis is not known.

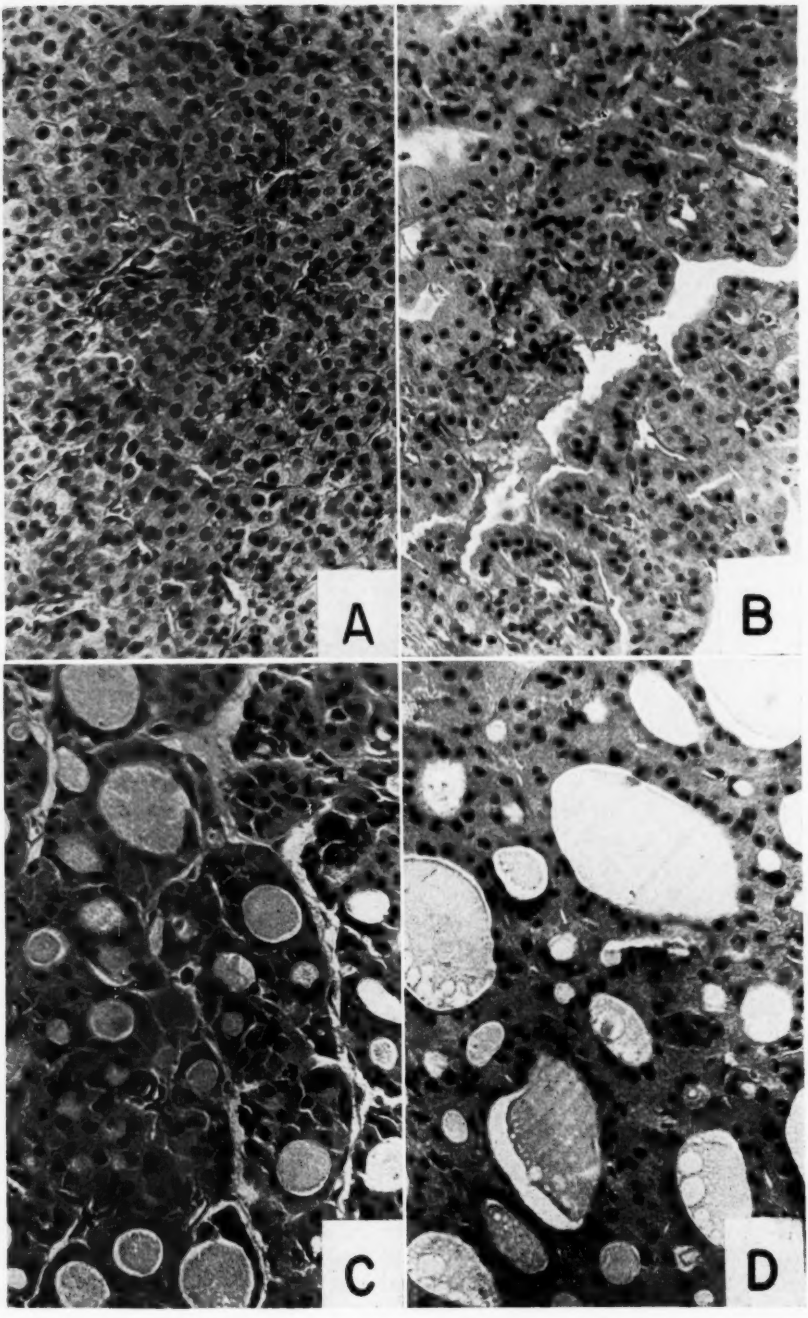
In no case in the present series was the diagnosis made before operation, and there was not even suspicion of cancer in 20 of the 22 patients. Chesky and his associates [1b] remark that in their series the preoperative diagnosis was a benign condition in 23 of 25 patients, while Frazell and Duffy [1d] noted that in 53% of their series of previously untreated patients no carcinoma was suspected before surgery.

Chesky and his associates [1b] are of the opinion that removal of the involved lobe without neck dissection will cure in most cases so long as there is no evidence of tumor outside the gland. If cervical nodes are palpated, the patient should, in their opinion, have total thyroidectomy, radical neck dissection, and postoperative external irradiation. Frazell and Duffy [1d] prefer total lobectomy for the solitary nodule and radical neck dissection in cases of residual or recurrent tumor. They report that in 17 of their 19 referred patients (previously treated) who had metastases, surgical treatment could be considered to have been definitive (subtotal thyroidectomy), and yet 14 of these patients had recurrence or residual tumor. Eight of the latter patients ultimately died of their disease. Positive nodes were found in 25% of their series of patients at some time, but none were found in six patients whose primary treatment included radical neck dissection following aspiration biopsy or frozen section diagnosis.

External irradiation has been of little value. In the series of Frazell and Duffy [1d] it was employed in seven patients who had recurrence, and little improvement was noted. Radioactive iodine has also been used in some cases, but has had definite limitations because the tumor does not usually concentrate the material. Fitzgerald and associates [14] reported radioactive iodine concentration in only 3 of 19 Hürthle cell carcinomas studied—13 primary tumors and 6 metastatic lesions.

Chesky and associates [1b] reported that 88% of their patients were alive one and a half to nine years postoperatively, and 80% of the total number were free of disease with local surgical treatment alone in 96% of the group. In one case node dissection was carried out on two occasions, in addition to local tumor excision. Frazell and Duffy [1d] reported 2 patients who died postoperatively, 12 patients

14. Fitzgerald, P. J.; Foote, F. W., Jr., and Hill, R. F.: Concentration of I^{131} in Thyroid Cancer, Shown by Radioautography: Study of 100 Consecutive Cases Showing Relation of Histological Structure to Function of the Thyroid Carcinoma, Cancer **3**:86-105, 1950.



Examples of various histological types of Hürthle cell carcinoma taken from present series. Classification is after Chesky and others.[1b] × 200. *A*, trabecular type. *B*, papillary type. *C*, alveolar type with some large follicles. *D*, large follicular type with some small alveoli.

who died of their disease, 4 patients living with tumor, and 4 patients dead of other causes without tumor. Eighteen patients survived without evidence of tumor. In the latter series local surgery was performed in 25 patients, with an additional node dissection in 10 of them. The remaining 15 patients were treated with palliative surgery or irradiation.

In the present series all patients had local surgical excision of the tumor as primary therapy. In addition one patient had radical neck dissection for recurrence, and also received external irradiation and radioiodine. This last patient is still alive with widespread disease six years after the initial therapy. Tables 2 and 3 summarize the present series.

From the experience in the present series it would appear that local surgical treatment of the tumor offers a good chance for cure. Excision of a nodule alone is probably not advisable because it is impossible to determine grossly whether the

TABLE 2.—*Treatment of Twenty-Two Cases of Hürthle Cell Carcinoma*

Excision solitary adenoma	7
Subtotal lobectomy	2
Total lobectomy	8
Total thyroidectomy	5
Radical neck dissection (in addition)	1

TABLE 3.—*End-Results in Twenty-Two Cases of Hürthle Cell Carcinoma*

Postoperative death	1
Dead with disease	1
Dead without disease (other cause)	1
Lost to follow-up	3
Alive with disease	1 *
Alive without disease	15 †

* Six years after diagnosis.
† Three patients less than 1 year postoperatively; 3 patients from 1 to 5 years postoperatively; 9 patients more than 5 years postoperatively (2 for 15 years, and 1 each for 17, 18, and 19 years).

lesion is malignant. Radical neck dissection is best reserved for treatment of recurrent or residual tumor, but should be carried out as a primary procedure along with thyroid excision if clinical judgment indicates that carcinoma is present.

SUMMARY AND CONCLUSIONS

Twenty-two cases of Hürthle cell carcinoma have been presented, with pertinent discussion concerning etiology, diagnosis, pathology, and treatment.

Hürthle cell carcinoma usually resembles a benign adenoma clinically, but it can metastasize widely and therefore should always be considered as malignant and treated accordingly. In only one case in the present series distant metastases developed.

From the experience in this series it is also clear that all solitary nodules of the thyroid should be surgically excised, preferably by lobectomy.

Radical neck dissection should be reserved for those cases in which there is clinical evidence of spread of the tumor, or in which there is residual or recurrent tumor.

External irradiation and radioactive iodine have not proved effective in treating Hürthle cell carcinoma.

UROLOGICAL COMPLICATIONS FOLLOWING ABDOMINOPERINEAL RESECTION OF THE RECTUM

GEORGE O. BAUMRUCKER, M.D.
AND
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CHICAGO

THE FOLLOWING review regarding the urological complications after abdominoperineal resection of the rectum or rectosigmoid is based on a review of 105 patients operated on. The authors have attempted to formulate a set of conclusions from this review which they hope may serve as an aid in preventing the urological complications most frequently associated with this operation. All resections of the rectum were done for carcinoma, except in two patients. One operation was performed on a paraplegic for severe ulcerative colitis and the other for an extensive benign polyposis of the rectum.

Two series of patients were studied. The first group was evaluated on the basis of a two-year postoperative follow-up examination. The second group was followed through completely from the preoperative and postoperative examinations over a one and one-half year postoperative period. In the first series, 49 patients came to surgery in the two-year period of 1947 and 1948. Of these, 16 died within this two-year period. In the second series, 56 patients came to surgery in the years 1949-1951, and, of these, seven died in the year and a half postoperative follow-up period. Of the two series of 105 patients, there were two preliminary ureteral implants into isolated segments of the ileum, or cecal pouch, done because of anticipated cystectomy. One patient died of homologous serum jaundice one month postoperatively. The other died of coronary thrombosis three weeks postoperatively.

The urological complications in this group of 105 patients were, in the order of frequency, as follows: bladder dysfunction in the form of postoperative urinary retention, ureteral injuries, and perineal fistula (Table 1). The most important complication from the standpoint of immediate postoperative care was the associated bladder dysfunction. The two other complications may be considered more or less matters of surgical technique, although all three were in the final analysis due directly or indirectly to the surgery. In considering the two less important complications first, it is to be noted that ureteral injuries can be troublesome and, if bilateral, can be fatal. They occur in an overall estimate of 6% of the patients operated on. In this series there were six ureteral injuries—three left, one right, and

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TABLE 1.—*Summary of Urologic Complications Following Abdominoperineal Resection of the Rectum*

Total no. of patients operated on for carcinoma of rectum	105
Ureteral injuries (3 left, 1 right, 1 bilateral)	6
Postoperative perineal urinary fistulae	6
Patients with no bladder dysfunction following removal of temporary postoperative catheter	84
Patients with persistent postoperative residual urine *	21
Associated prostatic hypertrophy †	17
Retention not associated with bladder-neck obstruction	4
Retention believed due to bladder pain ‡	2
Retention believed due to more permanent injury to parasympathetic nerve plexuses	2

* All showed some degree of hypotonic bladder.
† All relieved by transurethral resection.
‡ One relieved by chordotomy (pericystitis); one metastasis to perineum (not relieved).

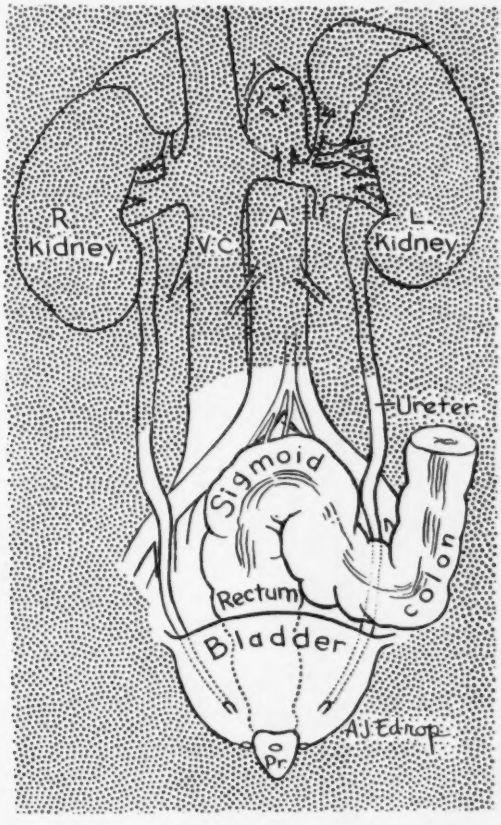

Fig. 1.—Drawing illustrates anatomical proximity of the sigmoid colon to the left ureter and demonstrates the reason for more frequent injuries to this ureter during removal of the recto-sigmoid.

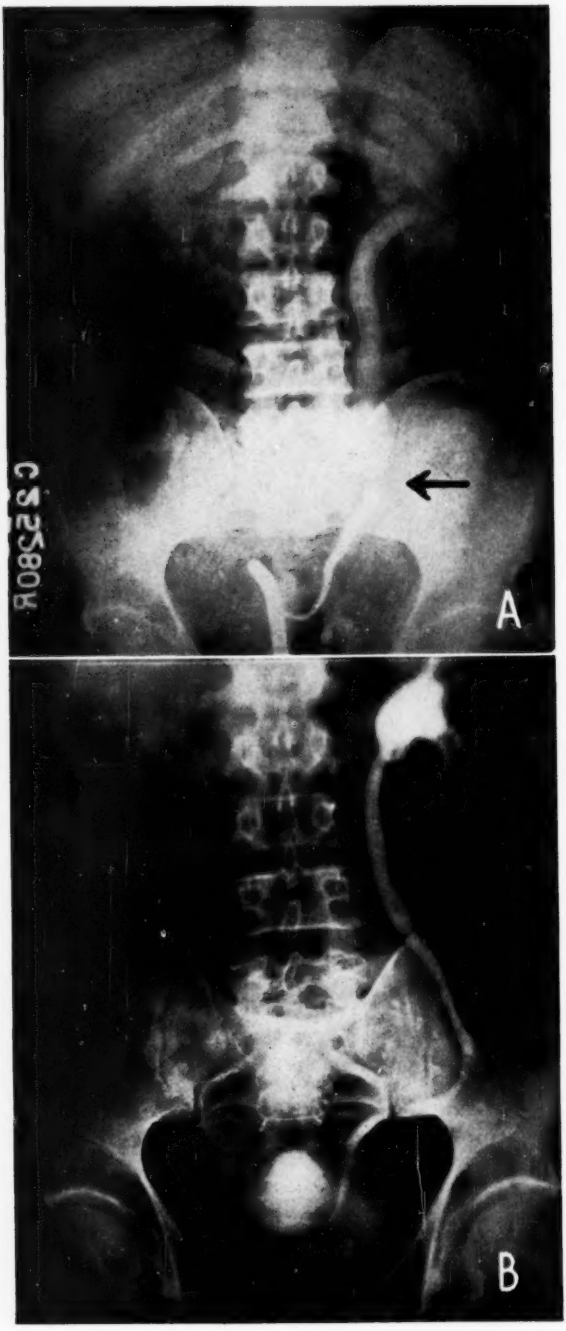

Fig. 2.—*A*, photograph shows postoperative ureterogram eight months following resection of the rectum. Arrow points to sharp block in the ureter, with dilatation above point of obstruction. *B*, retrograde ureterogram two months following corrective surgery of left ureter.

one bilateral. Figure 1 shows how both the lower loop of the descending colon and the ascending loop of the sigmoid can involve the left ureter, especially in the more extensive carcinomas, and offers a possible explanation for the preponderance of left-sided ureteral injuries. This percentage is based on known ureteral

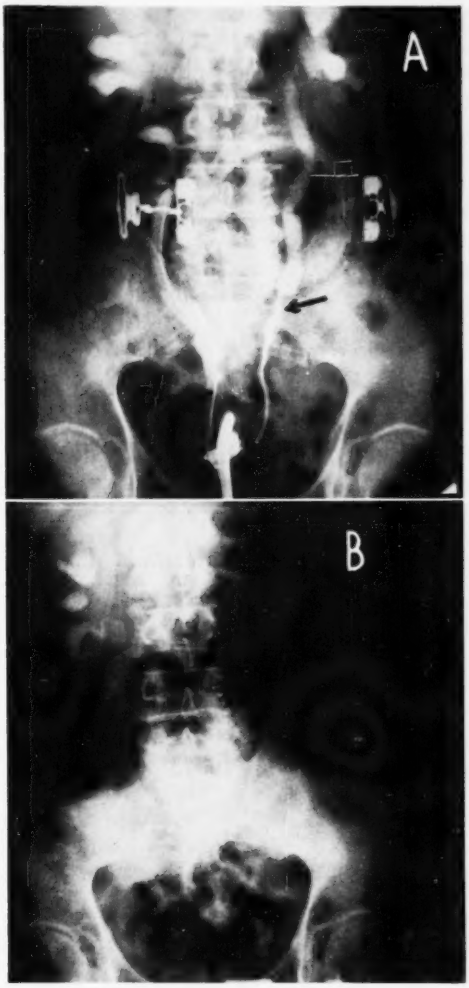

Fig. 3.—*A*, retrograde pyeloureterogram shows bilateral hydronephrosis, nonsymptomatic, one year following surgery of the rectum. Arrows point to the site of each ureteral constriction. *B*, postoperative intravenous pyelogram shows nonfunction of left kidney and hydronephrosis of right kidney. Preoperative intravenous pyelograms showed normal kidneys.

ligations at the time of operation and those discovered during the course of the postoperative follow-up examination. It is therefore very possible that the true percentage of ureteral injuries is much higher, since the injuries are usually done accidentally and may go unrecognized for months or even years. For example,

in Figure 2 the ureteral obstruction and nonfunctioning hydronephrotic left kidney seen here was discovered eight months following the operation. Examination was made because of vague left-sided backaches dating from the time of the operation. Retrograde pyelograms disclosed a completely blocked kidney with no function. One patient died of uremia nine days following the abdominoperineal resection, with progressive oliguria and increase in nonprotein nitrogen up to 240 mg. No autopsy was obtained, but it was believed that this was possibly a second bilateral ureteral injury or ligation not included in the above statistics.

Persistent postoperative perineal urinary drainage is not frequent but does occur and can complicate an otherwise smooth convalescence. It has been found in approximately 5% of the operative patients. In this series all six perineal fistulae closed after a varying time of Foley urethral catheter drainage. This condition is usually found in those patients in whom the rectal tumor has invaded the bladder and prostate. The carcinoma may involve the posterior bladder wall, necessitating a resection of part of the bladder, or it may have spread to the prostate and vesicles, making a clean resection difficult. When the bladder neck

TABLE 2.—*Age Dispersal of One Hundred Five Patients Having Abdominoperineal Resections of the Rectum for Carcinoma* *

Age, Yr.	No. of Patients
70-76	7
60-70	20
50-60	53
40-50	8
30-40	17
Total	105

* Seventy-eight per cent between 50 and 76 years of age (prostatic age).

is involved, a bilateral ureteral injury is a real danger in any extensive dissection (Fig. 3). This brings up the question of a preliminary urinary diversion in those cases in which, because of tumor invasion of the bladder, a total cystectomy may also be indicated in connection with the rectal resection and colostomy. The so-called artificial bladder, or cecal pouch, operation may have some advantages in this connection.

The most common complication of the operation was seen in the associated bladder dysfunction (Table 2). This manifested itself in the form of a complete urinary retention, a high residual urine, or a weak urinary stream, with dribbling in about 20% of the patients operated on. Of this group of 21 patients with persistent postoperative urinary retention, 17 were found to have bladder-neck obstruction in the form of prostatic hypertrophy or median bar fibrosis; 4 appeared to have a normal bladder neck. Pre- and post-operative cystometrograms (Fig. 4) showed a definite decrease in postoperative bladder tone in every patient who was sent for postoperative examination. However, we wish to state that, although 80% had no postoperative bladder symptoms, almost all patients, if examined by the cystometrogram, showed some decrease in the bladder tone postoperatively. This was only of a temporary or mild nature in almost all patients. The following symptoms were present in this group: decrease in active voluntary bladder pressure, increase in bladder capacity, and decrease in sensation of filling to fullness

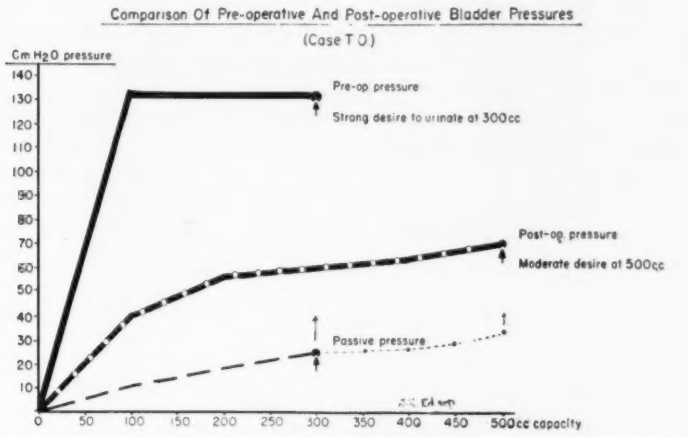

Fig. 4.—Graph showing comparison of pre- and post-operative bladder pressures. Patient could not exert more than 65 cm. of water pressure following the operation; whereas preoperatively he had no trouble in getting up to 130 cm. of water. Feeling of fullness was prolonged from 300 to 500 cc. capacity.

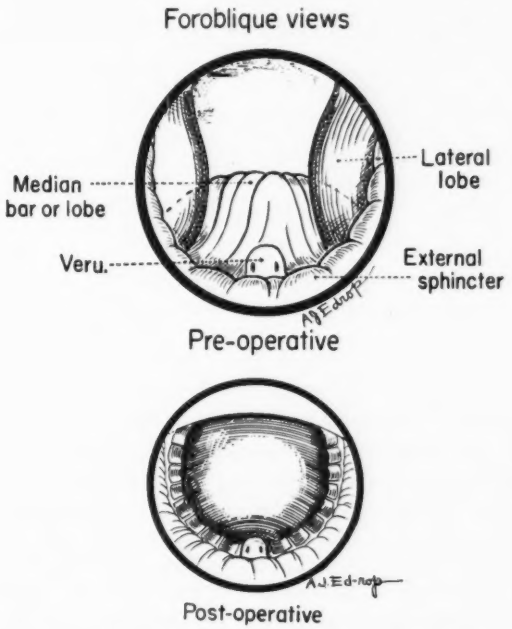

Fig. 5.—Cystoscopic (foroblique) view of the bladder neck of a moderate prostatic hypertrophy of the type most commonly found associated with urinary retention in these patients. This type of prostatic hypertrophy is many times nonsymptomatic and hence unrecognized preoperatively because of a certain amount of compensatory detrusor hypertrophy.

and a residual urine. For example, where preoperatively there was an active pressure of 80 to 90 cm. of water and a strong desire to urinate at 300 cc. capacity, there would be seen postoperatively an active pressure of only 40 to 60 cm. of water, with only a weak or moderate desire to urinate at 500 cc. capacity, usually associated with a decreased sensation to filling. The degree of hypotonicity in this example, in the absence of any bladder-neck obstruction, would not cause urinary retention in itself. It has been noted by some surgeons [2] that a careful separation of the rectum close to the bowel wall prevents damage to the autonomic plexus and would therefore tend to produce less trauma and hence less postoperative motor and sensory depression. Table 3 shows analysis of the results

TABLE 3.—*Results of Transurethral Resections in Relieving Urinary Retention Following Resection of the Rectum*

Age, Yr.	Preoperative Genitourinary Symptoms	Abdomino-perineal Resection	Postoperative Findings	Transurethral Resection	Amount Resected, Gm.	Results
56	None	11/ 5/47	Complete retention, low pressure, 15 mm. Hg at 100 cc.	12/11/47 Median bar and laterals	10	15 cc. residual, good stream
59	Nocturia 3 times, 50 cc. residual	6/18/47	200 cc. residual, weak stream, 15 mm. Hg at 100 cc.	9/18/47 Median bar and laterals	12	Not satisfactory, 200 cc. residual
				10/ 2/47 (recurrence)	5	20 cc. residual, excellent stream
54	Nocturia, no residual	3/29/48	300 cc. residual, weak stream, 8 mm. Hg at 50 cc., 15 mm. Hg at 100 cc.	5/24/48 Median bar and lateral lobes	12	3 cc. residual, good stream
34	None	11/ 4/47	300 cc. residual, weak stream, 450 cc. capacity with dulled sensation	No operation, metastases, median bar	..	.....
51	None	8/ 1/47	Dribbling stream, no residual, 400 cc. capacity with dulled sensation	9/16/48 Median bar	5	Good stream
61	None	4/ 1/47	100 cc. residual frequency and nocturia, recurrence of high residual	7/27/48 Median bar	2	No residual at first, increased to 200 cc.
				4/ 4/49	2	30 cc. residual
59	Nocturia 2 times	2/ 4/47	140 cc. residual, weak stream since operation	8/ 2/48 Spastic sphincter and lateral lobes	10	Postoperative stricture, weak stream, 100 cc. residual
				8/ 9/48	23	40 cc. residual
55	None	2/ 1/48	Nocturia 6 times, 150 cc. residual	10/ 9/48 Median bar	4	Good stream, no residual
56	Nocturia 2 to 3 times	1/19/47	35 cc. residual, small stream (weak)	9/ 9/48 Median bar and laterals	12	Good stream, no residual

Average resected 12.

of transurethral resection on the residual urine in the first series of patients undergoing operation. Almost all patients with urinary retention were improved following a conservative bladder-neck resection (Fig. 5).

It was therefore believed that in most instances the retention symptoms were enhanced or produced by a suddenly weakened bladder detrusor being superimposed on an already existing bladder-neck obstruction, which may have been compensated up to the time of the operation. The cause of a high residual urine in the absence of a bladder-neck obstruction in four patients in this series may be attributed to one or all of the following possibilities: (1) injury to bladder innervation, found especially in extensive dissections; (2) disturbance or distortion of bladder neck due to either involvement of the prostatic urethra by the tumor or a backdrop of the bladder into the rectal space, and/or (3) direct injury to the bladder muscles, with resulting postoperative edema and reflex retention of urine due to pain.

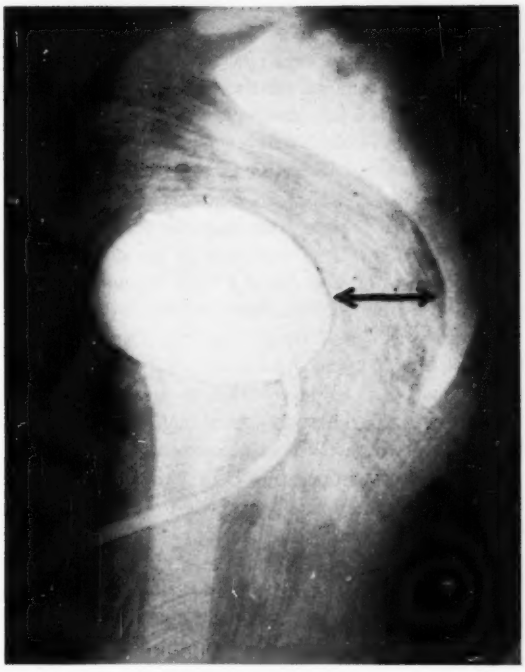

Fig. 6.—Lateral standing cystogram demonstrates relation between bladder and sacrum in four patients in whom postoperative urinary retention persisted, in spite of a normal and ample bladder neck. No backward displacement of the bladder could be demonstrated. Retention in these cases was believed to have been due to a truly hypotonic bladder.

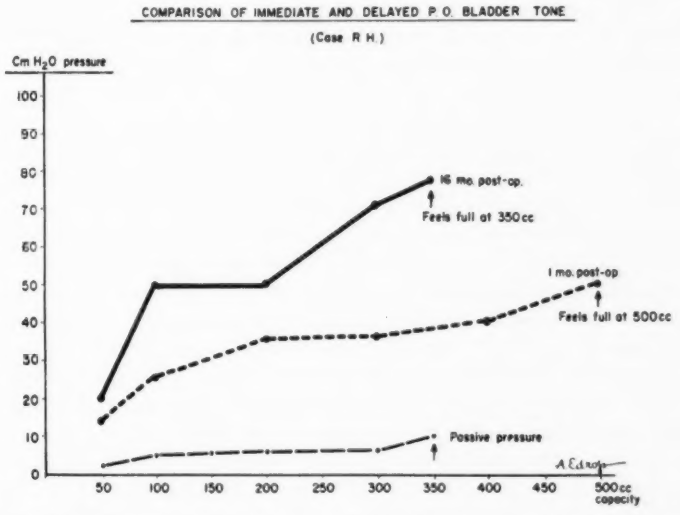

Fig. 7.—Graph shows comparison of immediate and delayed postoperative bladder pressures. In this patient there was a slow but definite recovery of bladder tone during the 16 months following surgery of the rectum.

Some authors [7] have noted that in an occasional patient the urinary retention set in immediately following removal of the perineal pack. This would seem to indicate that the pack supported the bladder and prevented a backdrop as long as it was in place. However, in this series of patients studied we were unable to discover any evidence showing a backdrop of the bladder into the space of operation or any distortion effects from such. Postoperative lateral standing cystograms made six months to one year postoperatively in these four patients who had persistent urinary retention with no evidence of prostatic hypertrophy showed (Fig. 6) a normal position of the bladder and no urethral constriction. The cause of retention in these four patients was divided between two truly hypotonic bladders due to severe parasympathetic injuries; one due to reflex retention caused by bladder pain (pericystitis?) and relieved up to the time of death by chordotomy, and the last one believed to have been due to pain of perineal metastases unrelieved by any measures. Although a severe drop of the basfond has not been noted in these patients, a moderate drop of the trigone, which has been seen immediately postoperatively in other patients, has been found to rise and level out on examination six months to one year following the operation. This may indicate that any original backward displacement that had occurred had a tendency to correct itself in time as the recuperative process filled in the rectal space. This may also be the explanation for the simultaneous return of bladder tone in the course of one year after the operation, as shown in Figure 7.

In a review of 25 consecutive abdominoperineal resections of the rectum [9] on women, it was found that in no instance was there any postoperative urinary retention which was not relieved by one or at the most two single postoperative catheterizations. No Foley retention catheters were used in this series. This might at first appear to support the contention that in a woman the vaginal and uterine support (not present in the male anatomy) prevents any backward sagging of the bladder. Although this may well be true, it is to be remembered that the incidence of a complicating bladder-neck obstruction in women is rare and that the urinary channel is short, direct, and of good caliber.

POSTOPERATIVE CARE OF THE BLADDER

A simple open system of drainage with a small type Foley catheter will bring these bladders back to a normal voiding in almost all instances. Comparison of the practices of different surgical services has shown a very significant relationship between the length of time of postoperative catheter drainage and the recurrence of postoperative urinary retention. Where the Foley catheter has been left in place two weeks following the operation, there were no instances of postoperative retention on removal of the catheter. On those services where the catheters were removed three or four days following the surgery, it was noticed that reinsertion of the catheter for recurrent retention was frequent. Bladders complicated by bladder-neck obstructions have in turn returned to a satisfactory voiding residual following transurethral resection in almost all instances.

The amount of the resected tissue in the prostates associated with retention in these patients has varied from 2 to 30 gm. In many instances only a small resection of 2 to 5 gm. was necessary to allow for the return of a good stream, increased force, and decreased residual urine. An occasional severer and more persistent type of hypotonic bladder would usually necessitate a longer period of post-

operative drainage. A parasympathetic stimulant such as bethanechol (Urecholine) chloride may have a useful though not frequent role in the treatment of these bladders in the absence of bladder-neck obstruction (Fig. 8). Several patients had a mild but definite sensory impairment manifested by an absence of a desire to urinate except with a distended bladder. With 5 gm. of bethanechol chloride only, two or three times a day, the desire to urinate has been maintained in the few patients on whom it was tried and appeared with smaller amounts of urine in the bladder.

CONCLUSIONS REGARDING PRE- AND POST-OPERATIVE RECOMMENDATIONS TO THE SURGEON

The presence of a hydronephrosis, or nonfunctioning kidney, should be determined preoperatively to allow for proper precautions to be taken. This may be a very important factor in the preservation of the patient's kidney function or his

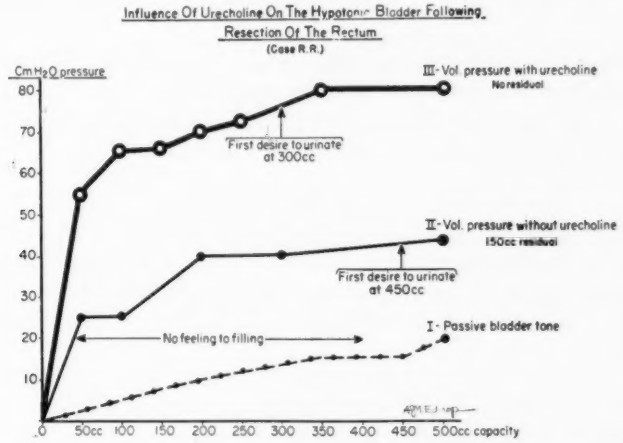

Fig. 8.—Graph showing effect of bethanechol (Urecholine) chloride on bladder pressure. Under bethanechol chloride medication, postoperative bladder pressure of 40 cm. of water was raised to a level of 80 cm. of water. The residual urine improved accordingly from 150 to 20 cc. under bethanechol chloride.

life. Death from a postoperative uremia has been observed by the authors nine days following resection of the rectum. Injuries to the ureter during surgery, as well as involvement of the ureter by the malignant process, have not been too uncommon in carcinoma of the rectosigmoid.[10] It is, therefore, wise to have a knowledge of the kidney function before surgery. Since lesions of the upper and lower sigmoid place the left ureter in jeopardy, it is most advisable to insert ureteral catheters before the operation to make the ureters easier to identify. The ureteral catheter also serves to make simpler the subsequent repair of a ureter that may have been inadvertently transected during the course of the operation and which may be deemed advisable or imperative to repair. On the other hand, rather than to chance a peritoneal contamination or a draining urinary fistula, which may easily result from an attempted ureteral repair, it may be wiser in some instances to completely tie off the ureter at the time of the operation and sacrifice the kidney, this, of course,

being done only if the remaining kidney is normal. This implies a preoperative knowledge of the function of each kidney. Therefore, the following set of rules was used as a guide for the prevention of the more serious postoperative complications:

1. Preoperative examination of the patient is done to determine the differential kidney function, bladder pressure, and status of the bladder neck. This can be completely covered by a preoperative intravenous pyelogram, rectal examination, and residual urine determination.

2. Preoperative transurethral resection of an obstructing bladder neck is done whenever such is present.

3. Preliminary insertion of ureteral catheters should be a routine procedure of this operation.

4. A urethral catheter is inserted postoperatively for at least 10 days to 2 weeks and is covered by suitable antibiotics to prevent cystitis.

5. Postoperative cystoscopic examination and bladder pressure study are done on those patients having a persistent urinary retention in order to determine its cause.

6. Transurethral resection is done on all in whom there is an associated prostatic hypertrophy not discovered preoperatively.

7. Intravenous pyelograms are made one to two months postoperatively to determine the final status of the ureters and kidneys.

SUMMARY

Ureteral injury and draining perineal fistulae occurred in approximately 5% of the patients operated on. Bladder dysfunction appeared in approximately 20% of patients subjected to combined abdominoperineal resection of the rectum. This dysfunction was manifested as complete retention, high residual urine, or weak urinary stream.

The bladder dysfunction is believed to be brought on by a mild or moderate degree of postoperative hypotonic bladder being superimposed on a previously existing bladder-neck obstruction in most cases.

The high incidence of associated prostatic hypertrophy in these patients can be explained by the similarity of the age group for both carcinoma of the rectum and prostatism.

While superficial damage to the parasympathetic nerves of the sacral and perivesical nerve plexuses may be an explanation for the temporary postoperative hypotonicity and urinary retention seen here, permanent damage to these plexuses, except as a rare exception, is not believed to be a very serious threat during this operation. The bladder tone returned sufficiently well during the period of postoperative catheter drainage in almost all patients.

In a small group (approximately 4%) the persistent urinary retention was believed to have been due to either (*a*) pain from a postoperative bladder edema, pericystitis, or perineal metastases, or (*b*) a more permanent damage to the parasympathetic fibers than is usually encountered. A chordotomy may relieve this first type of retention by relieving the pain. Parasympathetic stimulants, such as bethanechol (Urecholine) chloride or neostigmine (Prostigmin tablets) bromide, may help the latter type.

Definite relation seems to exist between the degree of postoperative bladder dysfunction and the extensiveness of the pelvic surgery.

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Advice and counsel was given by Dr. C. B. Puestow, Chief of Surgical Service, Veterans Administration Hospital, Hines, Ill.

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TRANSVERSE AND MUSCLE-SPLITTING ABDOMINAL INCISIONS

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A RECENT survey of over 500 certified surgeons in this country revealed that only about 26% are employing transverse or muscle-splitting abdominal incisions for the common types of abdominal operations.[1] It appears therefore that the majority of surgeons and most of the other physicians performing surgery are using midline or paramedian incisions. It seems inconceivable that adherence to these standard incisions by so many still exists, in view of the many unfavorable reports concerning the significantly high incidence of wound dehiscence and herniation which follows these incisions. These complications have been reported to occur in 2 to 10% of abdominal operations in which the midline or paramedian vertical incisions were employed,[2] whereas the reports of many surgeons using a muscle-splitting or transverse incision indicate that these two complications are rare, occuring in less than 1% of all cases.[3]

From the Department of Surgery, Veterans Administration Hospital.

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3. (*a*) Katz, I. A.: Early Ambulation in the Management of the Surgical Patient, Surg. Staff Meet., Veterans Admin. Hosp., Des Moines, Iowa **4**:1, 1950. (*b*) Leithauser, D. J.: Confinement to Bed for Only 24 Hours After Operation, Arch. Surg. **47**:203, 1943. (*c*) Leithauser, D. J.: Early Ambulation and Related Procedures in Surgical Management, Springfield, Ill., Charles C Thomas, Publisher, 1946, p. 93. (*d*) Mason, J. T.: New Abdominal Incision, Arch. Surg. **19**:129, 1929. (*e*) Middleton, E. E.: Transverse Abdominal Incisions, Texas J. Med. **46**:154, 1950. (*f*) Palumbo, L. T.: Esophagoduodenal Anastomosis in Selected Cases of Total Gastrectomy, J. Internat. Coll. Surgeons **14**:267, 1950. (*g*) Palumbo, L. T., and Brennan, J. E.: Results of Total Gastrectomy, ibid. **17**:823, 1952. (*h*) Palumbo, L. T.; Paul, R. E.; Katz, I. A., and Mazur, T. T.: Effects of Early Postoperative Ambulation, Postgrad. Med. **13**:206, 1953. (*i*) Pernworth, P.: The Transverse Incision in Abdominal Surgery, Am. J. Surg. **72**:573, 1946. (*j*) Sloan, G. A.: A New Upper Abdominal Incision, Surg., Gynec. & Obst. **45**:678, 1927. (*k*) Footnote 2*d*, *e*, *g*, and *h*.

Whitaker and his associates [2b] reported nine cases, 0.9%, of wound dehiscence occurring in 1,000 intra-abdominal (peritoneum was opened) operations in which a muscle-splitting or transverse incision was employed. Singleton and Blocker,[2e] in a survey of 5,853 operations (intra-abdominal) in which a vertical incision was used, noted that wound dehiscence occurred in 60 (1.02%) cases, whereas in 3,147 intra-abdominal operations in which a muscle-splitting or transverse incision was used, dehiscence occurred in only 1 case, an incidence of 0.032%. Mayo and Lee,[2c] in 20,300 intra-abdominal operations performed during the period 1945 to 1949, reported 94 cases, 0.45% in which a wound dehiscence developed. Of these 94 cases, 82 occurred when vertical incisions were used. Mayo stated that in the largest number of cases dehiscence followed a vertical rectus incision; a muscle-splitting rectus incision is widely used at the clinic.

The explanation for this difference is based upon sound anatomical and physiological principles.[4] A study of the nerve supply, the relational anatomy of muscle attachments with their directional pull, and the stronger directional components of the aponeurotic fibers are the main factors involved.

In long vertical incisions about three of the intercostal nerves supplying the rectus abdominis muscle are injured or divided, thereby producing muscle atrophy and weakness. In addition to this, and more important, the anterior and posterior rectus sheaths are divided vertically, thereby transecting the aponeurotic fibers. It is thus evident that in the closure of these sheaths the sutures are placed parallel to the direction of these strong fibers. This coupled with the normal lateral pull of the muscles of the anterior abdominal wall places severe tension upon the incision, which may result in the tearing out or pulling through of the sutures (Figs. 1 and 2). With use of a transverse incision, injury to nerve supply is unlikely; also, when this incision is made the division of the rectus sheaths is in a transverse direction parallel to the strong fibers so that in closure of these layers the sutures will pass across rather than parallel to these fibers, thereby making the closure more secure. This closure is kept in approximation by the lateral pull of the abdominal muscles, rather than pulling the line of incision apart as in a vertically placed incision. The same is true for muscle-splitting incisions. The lateral direction of muscle pull will keep the transverse incision in approximation (Figs. 1 and 2).

This mechanism of tension and pull paralleling the line of incision is also a favorable factor in reducing postoperative wound pain and discomfort. With less pain, the patient will cough more readily, move about in bed more freely, and be out of bed sooner. These increased activities during the early postoperative period will significantly reduce the incidence of postoperative complications.

The lateral pull by the muscles of the anterior abdominal wall upon a vertically placed incision was demonstrated by Sloan.[3j] He showed that the vertical strain and stress coefficients on the suture lines of these incisions and wounds are proportionate to the square of the length of the incision. This force is 30 times greater in a vertical incision than in a transverse incision; a 30 lb. (13.6 kg.) force is necessary to approximate a 3 in. (7.6 cm.) incision and an 80 lb. (36.3 kg.) force for a 5 in. (12.7 cm.) incision.

4. Kimbarovskiy, M. A.: Experimental Study on the Effect of Early Rising on Healing of Postoperative Wounds, Novy khir. arkhiv. **48**:116, 1940. Lampe, E. W.: Surgical Anatomy of the Abdominal Wall, S. Clin. North America **32**:545, 1952.

Sectioning one or both rectus abdominis muscles has no serious anatomic or physiologic consequence. There is no interference with the nerve or blood supply. The muscle does not retract for any distance, since it is prevented from doing so by the tendinous bands which are normally present, attaching it to the under-

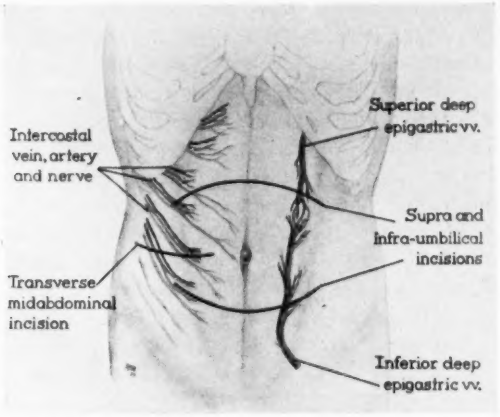
Superior deep epigastric vv.
Intercostal vein, artery and nerve
Supra and infra-umbilical incisions
Transverse midabdominal incision
Inferior deep epigastric vv.

Fig. 1.—Skin incisions in relation to bony landmarks and neurovascular supply.

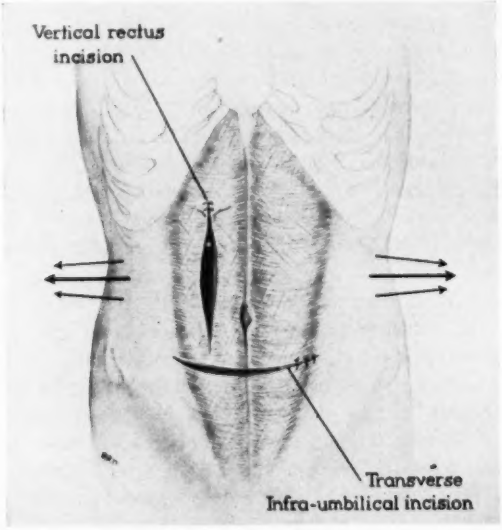

Fig. 2.—Relation of transverse and vertical incisions to the strong fibers of the rectus sheaths and pull of the muscles of the abdominal wall in the lateral direction, indicated by the arrows.

surface of the anterior rectus sheath. In the reapproximation of the divided rectus sheath, the rectus muscle is likewise approximated, and it requires no suturing (Fig. 3). The healing process results in the formation of another fibrous band, which does not impair the normal function of this muscle.

TYPES OF INCISIONS AND RESULTS

This report is based upon a follow-up study of only two of the various types of muscle-splitting or transverse abdominal incisions that we have employed during the past seven years. Included are 220 transverse elliptical supraumbilical and infraumbilical incisions and 273 transverse midabdominal incisions. The supraumbilical incision was employed for partial and total gastrectomies and for operations on the pancreas, transverse colon, duodenum, and upper small bowel.

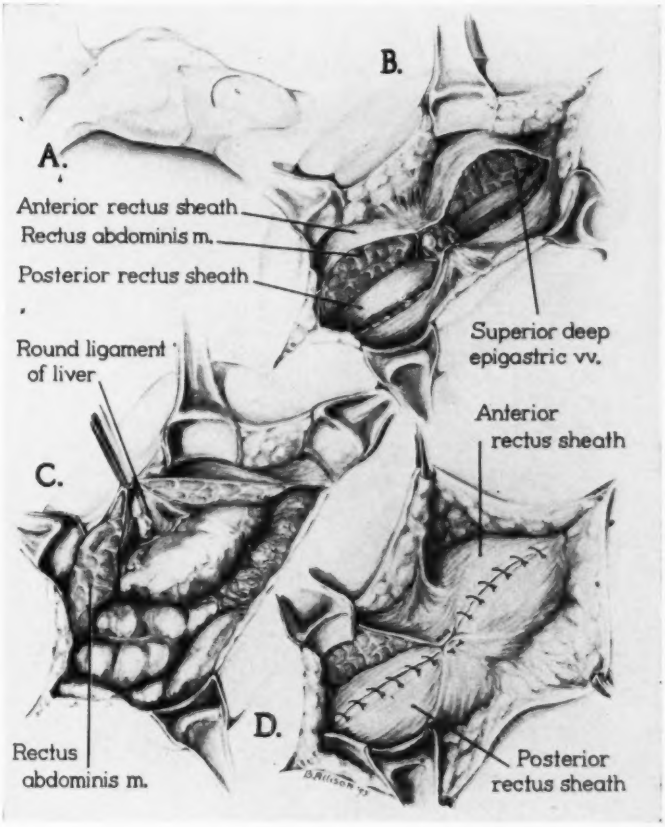

Fig. 3.—Transverse and surpraumbilical incision: *A*, skin incision; *B*, division of anterior rectus sheath and both rectus abdominis muscles; *C*, all layers of abdominal wall divided transversely, revealing some of the viscera of the upper abdominal cavity; *D*, closure of the posterior rectus sheath and partial closure of the anterior rectus sheath with interrupted 00 silk, using two mattress sutures in the midline.

The infraumbilical incision was utilized for total colectomies, small bowel surgery, and lesions in the pelvic cavity and retroperitoneal spaces; all these incisions were of the transabdominal (intraperitoneal) type. The right or left transverse midabdominal incision was employed for operations in the retroperitoneal spaces, for ligation of the inferior vena cava, for lumbar sympathectomies, and also for

hemicolectomies; in 236 cases in this group the operations were for lumbar sympathectomies, which were accomplished retroperitoneally without opening the peritoneum, whereas in the remaining 37 cases the operation was of the transabdominal (intraperitoneal) type. Of the latter group, 17 were for hemicolectomies, 6 for resection of the terminal ileum, and 14 for ileotransverse colostomies.

Supraumbilical or Infraumbilical Incisions.—The elliptical supraumbilical or infraumbilical incisions were performed by dividing transversely the anterior and

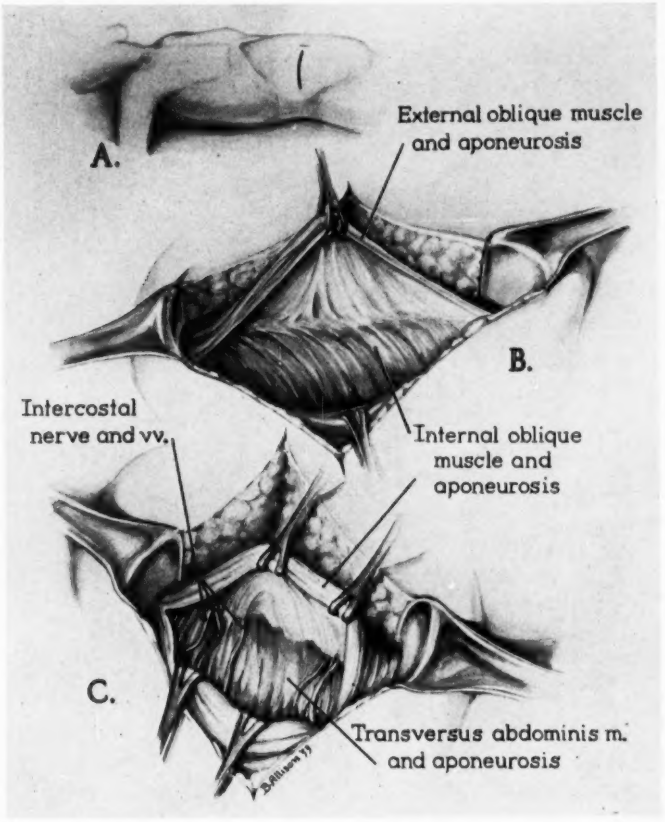

Fig. 4.—Transverse midabdominal incision: *A*, skin incision; *B*, external oblique muscle and aponeurosis divided in direction of its fibers; *C*, internal oblique muscle divided in direction of its fibers.

posterior rectus sheaths and transecting both rectus abdominis muscles (Fig. 3*A* and *B*). The deep superior and/or inferior epigastric vessels, as the case may be, were ligated and divided. The peritoneum was opened in the same direction, along with the posterior sheath (Fig. 3*C*).

The closure of the incision was accomplished with interrupted 00 nonabsorbable surgical (silk) sutures in the anterior and the posterior rectus sheath layers and 000 silk in the subcutaneous and skin layers (Fig. 3*D*).

Age Incidence: The average age of the patients in the supraumbilical group was 48 years and in the infraumbilical group, 32 years. In both series over 57% of the patients were ambulatory within 24 hours, whereas the remaining number were ambulatory after the sixth postoperative day.

Wound Complications: Dehiscence and incisional hernia did not occur in this series. There were only two wound complications, one infection and one hematoma.

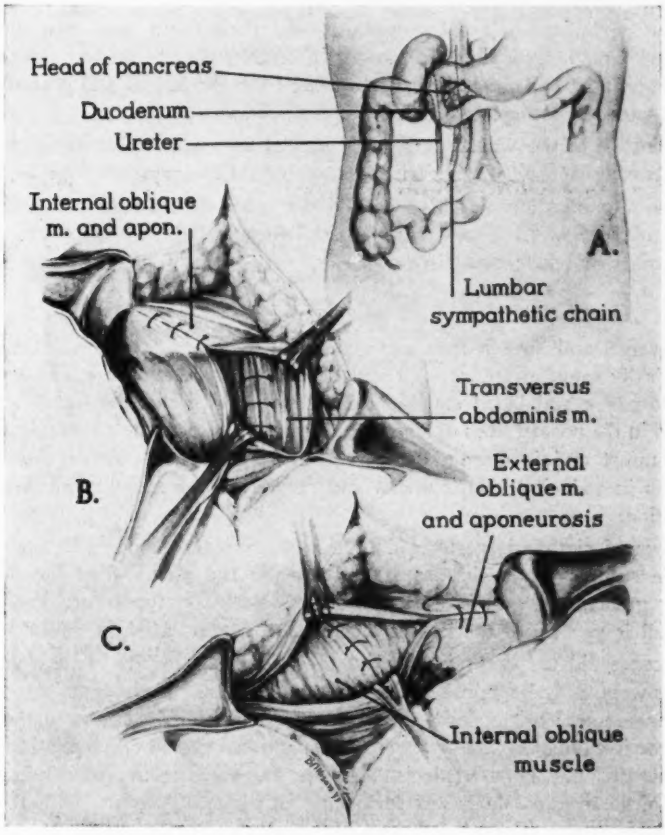

Fig. 5.—Transverse midabdominal incision: *A*, some of the structures and organs which can be exposed through this incision; *B*, suturing of the transversus abdominis and internal oblique muscles with 00 silk; *C*, suturing of the internal and external oblique muscles with 00 silk.

Transverse Muscle-Splitting Incisions.—The transverse midabdominal incision was performed in 273 cases. Of this number, 236 were for a right or left lumbar sympathectomy, 17 for hemicolectomy, 6 for resection of the terminal ileum, and 14 for ileotransverse colostomy.

Age Incidence: The average age of the patients in this group was 49 years; 78% were ambulatory within 24 hours.

Operative Technique: The skin incision was transverse, placed at the level of the umbilicus; the medial edge began about 3 in. (7.6 cm.) from the midline and extended lateralward (Fig. 4*A*). The skin flaps were mobilized superiorly and inferiorly in order to expose a broad expanse of the underlying muscles of the anterior abdominal wall. The external oblique aponeurosis and muscle were split in the direction of their fibers and separated from the underlying internal oblique muscle (Fig. 4*B*). This lateral structure and the transversus abdominis muscle were likewise split in the direction of their fibers (Fig. 4*C*). If the operation to be performed was intraperitoneal, the peritoneum was then incised in the vertical direction. If a retroperitoneal procedure was to be accomplished, the extraperitoneal fat and peritoneum were freed from the lateral and posterior body wall and were then displaced upward and medialward.

The closure of the wound was accomplished by reapproximating each muscle and aponeurotic layer separately with interrupted 00 silk sutures (Fig. 5*B* and *C*).

Wound Complications: Dehiscence did not occur in this series. A small incisional hernia developed in one case, an incidence of 0.37%. Infection occurred in three cases and hematoma in two cases.

SUMMARY

Transverse and muscle-splitting abdominal incisions are anatomically and physiologically sound procedures. They provide adequate exposure, cause minimal damage to nerve and blood supply of the structures of the abdominal wall, and are placed in the normal lines of stress and strain. Thereby, the patient experiences less discomfort and is more cooperative during the early postoperative period, which aids in reducing complications and results in more rapid ambulation and rehabilitation.

The wound closure is performed by a multilayer technique, with use of nonabsorbable sutures across the normal aponeurotic and muscle-fiber bundles, thus providing greater strength in closure and less strain on the suture line because muscle pull is parallel to the line of incision rather than at right angles to it.

Dehiscence did not occur in a series of 497 cases. In 261 of these cases the operations were transabdominal (intraperitoneal) in type. In the remaining number the peritoneum was not opened. There were no incisional hernias in the supraumbilical or infraumbilical groups. A small incisional hernia developed in one case in the transverse midabdominal incision group; this occurred in a patient in whom a resection of the terminal ileum was performed for regional enteritis. This represents an over-all incidence of 0.2% for the entire series and an incidence of 0.38% for the series in which a transabdominal (intraperitoneal) incision was employed. These two important complications occur infrequently as compared with those following vertically placed abdominal incisions.

DIFFERENTIAL HYPOTHERMIA FOR INTRACARDIAC SURGERY

Preliminary Report of a Pump-Oxygenator Incorporating a Heat Exchanger

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A SOUND technique for intracardiac surgery requires direct-vision access to the entire interior of the heart for prolonged periods plus adequate protection for the brain and other vital organs during cardiac isolation or failure. This cannot easily be provided at normal temperatures because of the technical difficulties presently inherent in large capacity extracorporeal pump-oxygenators. The protective action of reduced metabolism provided by hypothermia is but a partial solution to the problem.[1] Combined with a low capacity pump-oxygenator, however, hypothermia provides safety during lengthy cardiac isolation or failure. Direct cooling or warming of the blood is much simpler than external heat exchange, and in addition provides a saving in time and total caloric transfer, the heart and presumably other vital organs being cooled much more rapidly than the animal as a whole.

METHOD

Large mongrel dogs were lightly anesthetized with alpha chloralose, heparinized, and arranged as in Figure 1. The fenestrated femoral vein cannula measured 4 to 6 mm. in diameter, and was placed in the inferior cava as high as the diaphragm. The carotid artery cannula measured 3 mm. in diameter. Tubing and connectors were of polyethylene or polyvinyl plastic. The refrigerator coil was copper. The diaphragm pump, used at a fraction of its capacity, has been described.[2] The extracorporeal circuit was not siliconed and was filled only with normal saline, its volume being about 500 cc. The homologous lung was removed fresh and was cannulated under water to exclude air. Venous blood under a hydrostatic pressure of about 50 cm. of water was supplied to the lung via a cannula in the pulmonary artery. Oxygenated blood was collected from the left auricle by means of a cannula secured at the mitral ring.

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Resident in Surgery, Public Health Service Hospital (Dr. Polley).

Section on Technical Development, National Heart Institute (Dr. Bowman).

From the National Heart Institute, National Institutes of Health, Public Health Service, Department of Health, Education, and Welfare.

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2. Southworth, J. L.; Peirce, E. C., II; Tyson, T., and Bowman, R. L.: Diaphragm Pump for Artificial Circulation: Tyson-Bowman Pump, Arch. Surg. **66**:53, 1953.

An effort was made to maintain external blood flow at about 500 cc. per minute. Cooling was carried out until the heart reached 20 C. (68 F.). A curarizing agent was given occasionally to facilitate cooling when shivering became excessive. Rectal and heart temperatures were allowed to stabilize before warming to 32 C. (89.6 F.) was undertaken by changing the ice mixture for water at 40 C. (104 F.). After rewarming, the animals were killed. The following data were recorded: pericardial, rectal, venous outflow (femoral), and arterial inflow (carotid) temperatures; arterial and venous pressures; electrocardiogram; and blood flow in external circuit. Plasma hemochromogen was measured in some instances.[3] When fibrillation occurred, it was not interrupted for approximately 30 minutes, reversion then being effected with electric shock. About 500 cc. of blood was added during each experiment, and when blood pressure fell to dangerous levels in spite of this small doses of phenylephrine (Neo-Synephrine) hydrochloride (0.1 to 0.3 mg.) were given.

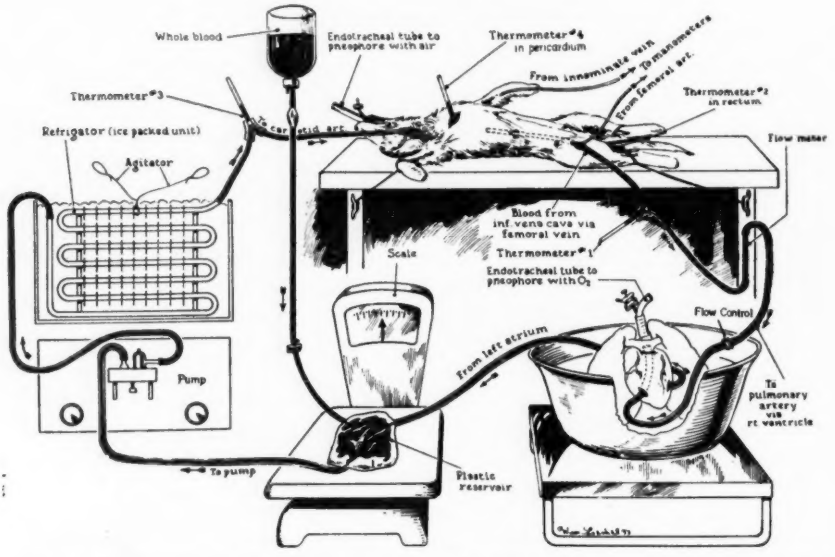

Fig. 1.—Semidiagrammatic representation of animal and apparatus. Flow is controlled by a screw clamp regulating venous inflow into the lung. Pump action is adjusted so that the weight of blood in the plastic reservoir remains constant. Flows are determined gravimetrically by clamping the inflow or outflow of the reservoir for 5 to 15 seconds. These flows are used to standardize the rotameter, which is then read for intermediate points.

RESULTS

Cooling experiments were carried out on 12 dogs, temperature and flow data of the last 8 being suitable for analysis (Table). Despite low flow rates in several experiments, cooling of the heart to 20 C. was effected in an average of 21 minutes. The rectal temperature lagged 5 degrees (C.) or more behind the heart temperature, and the total caloric exchange was only 34% of that calculated to cool the entire dog to 20 C. An average of 6.2 kg.-cal. per minute was lost during cooling with a flow of 420 cc. per minute. A fairly sharp rise in heart temperature to about 25 C. (77 F.) occurred on cessation of cooling. The rectal temperature also often rose

3. Hunter, F. T.; Grove-Rasmussen, M., and Soutter, L.: A Spectrophotometric Method for Quantitating Hemoglobin in Plasma of Serum, Am. J. Clin. Path. **20**:429, 1950.

*Data from Eight Consecutive Experiments**

Dog No.	Weight, Kg.	Average Flow, Cc. Per Min.	Min. to Cool Heart to 20 C.	Kg.-Cal. Lost Per Min. in Cooling	Calculated Kg.-Cal. to Cool Whole Dog to 20 C.	Measured Kg.-Cal. to Cool Heart to 20 C.	Per Cent of Calculated Calories to Cool	Min. to Warm to 32 C.	Kg.-Cal. Gained Per Min.	Kg.-Cal. Gained in Warming
P-2-52	17	200	14	3.9	240	55	23	37	1.0	40
P-83-53	28.5	575	21	6.1	400	130	32	32	2.1	65
P-104-53	27	210	30	4.0	405	120	30	29	1.6	45
P-98-53	18.5	555	11	8.9	285	100	35	25	2.0	50
P-68-53	21	400	21	6.3	360	130	37	18	2.4	45
P-125-53	18.5	480	11	7.0	280	80	28	18	2.9	50
P-130-53	21	520	15	8.7	320	130	39	15	2.8	40
P-135-53	28.5	405	41	4.6	485	185	39	43	2.9	125
Average	22.5	420	21	6.2	345	115	34	27	2.2	60

* Minute caloric exchange indicates much greater efficiency during cooling than during warming. Cardiac temperature is brought to 20 C. with only a fraction of the calories necessary to cool the entire animal. Warming is incomplete, about 50% of the extracted calories being added to bring the temperature to 32 C.

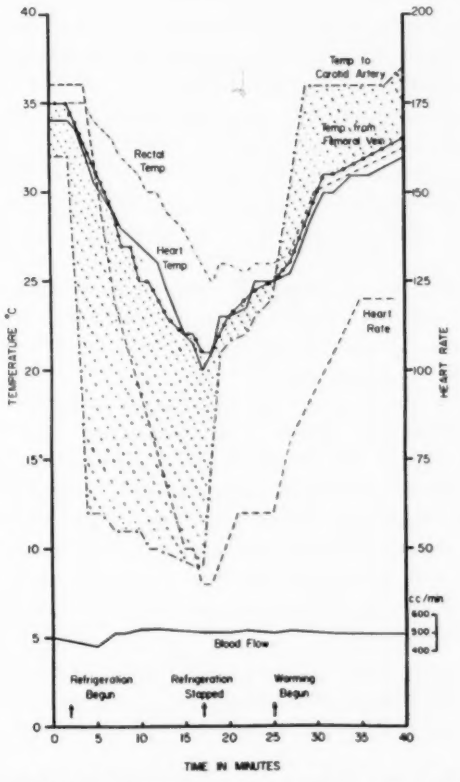

Fig. 2 (Dog No. P-130-53).—Temperature, flow, and heart rate data. The magnitude of heat exchange is represented by the area between the femoral vein and carotid artery temperature curves. Note that the temperature gradient is much greater during cooling than during warming and that the rectal temperature lags behind the heart temperature.

for several degrees. Warming, therefore, was begun at a heart temperature of about 25 C. It was satisfactory to about 32 C., though less than half as efficient as cooling, an average of 2.2 Cal. being exchanged per minute. Figure 2 illustrates the temperature relationships in a typical experiment.

Physiological changes during cooling and rewarming were generally similar to those reported by others except that venous pressure did not tend to rise.[1] Arterial pressure and heart rate fell markedly, and the component phases of the electrocardiogram were prolonged. The dog maintained some reflex activity even at the lowest temperatures and was quite responsive to stimuli after rewarming. Oxygenation appeared excellent as judged by color of arterial and venous blood. Hemolysis was not excessive, considering the extent of the unsiliconed surfaces, measuring 27, 42, and 157 mg. per 100 cc. at the completion of the experiment in three instances.

More than 500 cc. of blood flow per minute was generally obtainable by loosing the clamp on the venous inflow to the lung. Flow was sometimes impeded, however, because of intermittent obstruction of the cannula by the vein wall. This can probably be avoided by reducing the venous suction and by incorporating an inflatable balloon into the cannula tip.[4] Occasionally near the end of the experiment severe pulmonary edema developed in the homologous lung, and this markedly decreased blood flow by raising the pulmonary resistance. Ventricular fibrillation occurred three times in the 12 experiments. The arrhythmia was allowed to continue for about 30 minutes and then was reverted with one or more electric shocks before rewarming. Blood pressure was well supported during fibrillation in the cooled animal and rewarming was carried out in the usual fashion with no untoward event.

COMMENT

Differential cooling is a striking effect of directly refrigerating the circulating blood of a dog. The rate of cooling of any portion of the animal is probably proportional to the blood flow if all of the dog's circulating blood is refrigerated. This differential effect seems to be accentuated by cooling only a portion of the blood. For example, with a 500-cc. per minute flow of refrigerated blood from femoral vein to artery it was possible to reduce the rectal temperature to 13 C. (55.4 F.) while the esophageal temperature was still 34 C. (93.2 F.). The net cooling effect therefore depends both on organ flow and on where the blood is pumped in. When an artery in the cephalic end of the dog is used, it is probable that all vital organs will be cooled, while any differential effect due to partial flow will result in relatively greater cooling of heart and brain than of other organs. The details, of course, remain to be worked out. Warming of circulating blood showed no definite differential effect in these experiments.

Direct heat exchange involving externally circulating blood has been carried out by others, but the differential nature of the cooling seems not to have been considered. Gollan [5] effected cooling and warming of the entire circulating blood volume,

4. Gollan, F.; Blos, P., and Schuman, H.: Exclusion of Heart and Lungs from Circulation in the Hypothermic, Closed-Chest Dog by Means of a Pump-Oxygenator, J. Appl. Physiol. **5**:180, 1952.

5. Gollan, F.; Blos, P., and Schuman, H.: Studies on Hypothermia by Means of a Pump-Oxygenator, Am. J. Physiol. **171**:331, 1952.

using a simple silver coil in a pump-oxygenator circuit. Delorme [6] cooled blood circulating through a long external femoral artery to a femoral vein fistula made of plastic tubing. Blood was cooled to 10 to 20 C., and the temperature in the muscle of the thigh reached 22 to 29 C. (71.6 to 84.2 F.) in 30 to 45 minutes. No difference was noted by any of these workers in the cooling rate of different portions of the animals.

All experiments were acute since primarily temperature and caloric data were desired and problems of sterility, hemolysis, and so forth could be avoided. A rewarming temperature of 32 C. was selected because at this temperature the dogs can readily carry on without mechanical respiration and supportive drugs. Further rewarming can be effected by placing them in a warm room or heated blanket.

Oxygenation of blood appears satisfactory even at reduced temperatures. Although the diffusion rates of blood gases are slower, the affinity of hemoglobin for oxygen is increased, saturation being reached at a lower partial pressure.[7]

Homologous lungs were used here simply for convenience.[8] They were satisfactory except for the occasional occurrence of severe pulmonary edema. There is a choice of several good oxygenators for more permanent apparatus.

Ventricular fibrillation resulted in only 25% of the experiments, though all hearts were cooled to 20 C. It was always reversible by electric shock. The low incidence and easy reversibility may be related to maintenance of normal venous pressure, adequate cardiac oxygenation, or some other factor such as stable plasma pH.[9]

The details of temperature, blood flow, and oxygenation, in brain, kidney, liver, and other vital organs remain to be studied before this method can be considered suitable for use in humans. These experiments, however, strongly suggest that it will have practical application not only in intracardiac surgery but also in the treatment of certain types of asphyxia and shock.

SUMMARY

Experiments in large dogs using a low capacity pump-oxygenator incorporating a heat exchanger are described. With flows of about 500 cc. per minute the heart was cooled to 20 C. in an average of 21 minutes. Caloric exchange was only 34% of that calculated to cool the entire dog. Ventricular fibrillation was well tolerated and reversible. Warming was practicable to 32 C. by direct heating of the blood. The differential nature of the cooling process is discussed.

Alfred G. Casper assisted in this project.

6. Delorme, E. J.: Hypothermia in Dogs, Technical Report ONRL-83-53, Office of Naval Research, London, June 22, 1953.

7. Brown, W. E. L., and Hill, A. V.: The Oxygen Dissociation Curve of Blood: Its Thermodynamic Basis, Proc. Roy. Soc. **94**:297, 1923.

8. Potts, W. J.; Riker, W. L.; DeBord, R., and Andrews, C. E.: Maintenance of Life by Homologous Lungs and Mechanical Circulation, Surgery **31**:161, 1952.

9. Ripstein, C. B.: Personal communication to the authors.

ADEQUATE BREAST BIOPSY

Experience in One Thousand Consecutive Personal Cases

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EARLIER diagnosis of breast cancer demands constant reconsideration and expansion of all available measures. Periodic self-examination of the breast, coupled with periodic examination by the physician, is the established first step. In the next step, the clinical evaluation of the lesion with respect to biopsy, the physician is confronted with the temptation of "watchful waiting." This pitfall can be overcome only by prompt biopsy of any area in the breast that varies from the

TABLE 1.—*Histologic Diagnoses in 1,000 Consecutive Breast Biopsies*

Benign (740 Cases, 74%)			Malignant (260 Cases, 26%)		
Lesion	No.	%	Lesion	No.	%
Cystic hyperplasia	619	61.9	Carcinoma	256	25.6
Fibroadenoma	86	8.6	Scirrhous	186	
Duct papilloma	31	3.1	Medullary	45	
Gynecomastia	29	2.9	Duct cell*	18	
Lipoma	12	1.2	Mucinous	7	
Fibrofatty tissue	8	0.8	Sarcoma	4	0.4
Fat necrosis	3	0.3	Fibrosarcoma	2	
Tuberculosis	2	0.2	Cystosarcoma phyllodes	1	
Plasma cell mastitis	2	0.2	Hodgkin's disease	1	
Supernumerary breasts	2	0.2			

* Three of these patients had Paget's disease.

surrounding tissue. It is the purpose of this paper to discuss our concept of adequate breast biopsy, its indications, its technique, and the results obtained in 1,000 consecutive personal cases during the past 10 years.

INDICATIONS FOR BIOPSY

Our concept of adequate breast biopsy is one in which sufficient tissue is removed to reveal the presence of any obvious or occult cancer by immediate thorough pathologic examination (Table 1). Despite the easy accessibility of the breast to biopsy, the cancer area may be missed by the surgeon or the pathologist. Over the years the senior author has been led to widen the area of excision, since in numerous instances a cancer was discovered at the periphery of the specimen,

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adjacent to a more obvious benign lesion, such as fibroadenoma or cyst, or was found to be cancer in what clinically was diagnosed as a benign lesion. In the present series this unpleasant surprise was encountered in 13.5% of the cancer group or 3.5% of all biopsies.

While the extent of the breast biopsy must be adequate to reveal the nature of the lesion, it should not needlessly mutilate the breast from the functional or cosmetic standpoint. In making an all-out effort to rule out cancer, it is evident that a young woman with a single small lesion would not require as wide an excision

FIGURE 1.—Relationship of Malignancy to the Age of the Patient

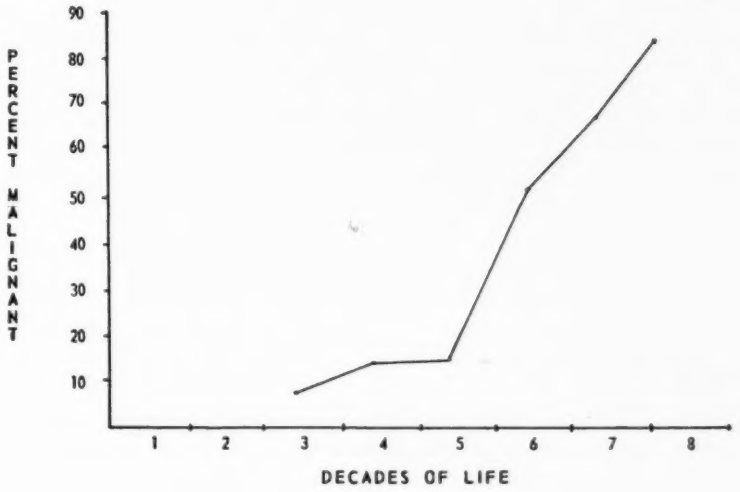


TABLE 2.—*Relationship of Tumor Size to Malignancy*

Size	% Malignant
Less than 1 cm. (small)	8.7
1 to 4 cm. (medium)	24.0
Over 4 cm. (large)	65.0

as an older woman with a less well-defined lesion (Fig. 1). It is also evident that in a large clinically obvious cancer a small incisional specimen for biopsy would be sufficient for establishing the diagnosis prior to radical mastectomy. In general the extent of the specimen for biopsy varies primarily with the type and size of the lesion (Table 2). Of less importance are the age of the patient and marital status, particularly with respect to childbearing. These factors are often so intimately related that they may not be clearly separable.

Cystic Hyperplasia.—The greatest confusion and uncertainty exists with respect to cystic hyperplasia in its various manifestations and its possible relationship to cancer. We have long abandoned the dangerous policy of "watchful waiting" in those patients thought to have cystic hyperplasia in whom there is a discrete area

that varies in consistency from the surrounding breast tissue. The presence of tenderness or pain over the lump or diffusely throughout the breast, as characteristically seen in cystic hyperplasia, does not reassure against a malignant lesion. In this series 22.4% of the patients with cancer presented this finding. While we can offer no proof of any etiological relationship between cystic hyperplasia and cancer, we have repeatedly seen these two lesions simulate each other clinically (25.4% of the cancers in this series) and have repeatedly seen them coexist in the same breast at operation. While the pathologist has reported the presence of cystic hyperplasia in only 15.4% of our cancer cases (Table 3), our gross surgical observations have revealed a much higher incidence. This is due to the fact that in large obvious cancers the pathologist frequently fails to report the incidental finding of cystic hyperplasia.

Conservatism pervades many quarters of medical thinking with respect to the diagnosis and management of cystic hyperplasia. Could one be sure that the lesion in question is only cystic hyperplasia and that cancer is excluded, there would be no quarrel with this policy. We have already condemned "watchful waiting." Aspiration biopsy and small incisional biopsy we feel are definitely inadequate to reveal the small or early carcinoma which may be harbored in an area adjacent to a more

TABLE 3.—*Lesions Associated with Carcinoma*

Lesion	No.	%
Cystic hyperplasia	41	15.4
Duct papilloma	5	1.9
Fibroadenoma	2	0.8
Lipoma	1	0.4

obvious lesion (13.5% of the cancers in our series). Furthermore, the reassurance from such a biopsy may be misleading to the surgeon and disastrous to the patient. We have had patients referred to us with cancer in whom these types of biopsy had been done so recently that it was evident the cancer area had been missed. Because of the frequency with which cancer may masquerade as cystic hyperplasia or be associated with it in a nearby area (Fig. 2), we strongly feel that only by removing the entire clinical lesion with a wide zone of surrounding, apparently normal breast tissue can the biopsy be deemed adequate. When the major duct system is widely dilated and filled with secretion, particularly in older women, we do not hesitate to remove it also.

Discharge from Nipple.—Patients with discharge from the nipple impose a special diagnostic problem. Causative lesions are duct papilloma, cystic hyperplasia, and carcinoma. Tables 4 and 5 give the percentage of these lesions in this series, as well as the type of discharge encountered. Although discharge from the nipple is relatively commonest in duct papilloma, cystic hyperplasia accounts for most cases because of its much greater clinical incidence. In our series 16.7% of the patients with duct papilloma had nipple discharge but accounted for only 10% of the total patients with discharge. On the other hand, 6.9% of the patients with cystic hyperplasia had nipple discharge but accounted for 71.7% of patients with discharge. The remaining 18.3% of patients had cancer associated with the discharge from the nipple. These experiences have led us to regard all patients with

any type of discharge from the nipple as cancer suspects, regardless of whether a mass is palpable or not. In one-third of our cases of duct papilloma the lesions were multiple. Herbut [1] from our own institution, states that 50% are multiple. We have come to regard duct papilloma as a potentially multiple lesion and therefore are not satisfied in removing single lesions.

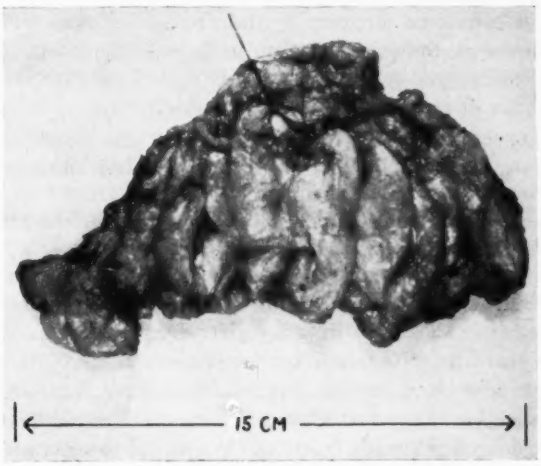


Fig. 2.—Specimen showing wide excisional biopsy in a case of cystic hyperplasia. Identification suture is placed at a small suspicious area which on frozen section proved to be carcinoma.

TABLE 4.—*Causes of Discharge from the Nipple*

Cause	No. of Cases	Relative %	% Cause of Discharge
Cystic hyperplasia (619 cases)	43	6.9	71.7
Duct papilloma (31 cases)	6	16.7	10.0
Carcinoma (256 cases)	11	4.3	18.3

TABLE 5.—*Types of Discharge from the Nipple*

Cause	No. of Cases		
	Serous	Bloody	Milk-Like
Cystic hyperplasia	17	21	5
Duct papilloma	2	4	..
Carcinoma	3	8	..

Because of the potential multiplicity of duct papillomas, because of the confusion which cystic hyperplasia presents with respect to nipple discharge, it is mandatory that in such patients the biopsy be adequate to rule out cancer within a wide zone of the major duct area. It is in this type of diagnostic problem that we perform our widest biopsies. This specimen for biopsy consists of a large core of breast tissue. The apex of the specimen for biopsy contains the entire major duct

1. Herbut, P. A.: Surgical Pathology, Philadelphia, Lea & Febiger, 1948.

system which is dissected from the areola, and the base includes the pectoral sheath, associated with one-fourth or more of the entire breast.

Fibroadenoma and Sarcoma.—When the lesion is a discrete, freely movable mass thought clinically to be a fibroadenoma, it usually suffices to remove the entire lesion with a surrounding zone of normal breast tissue. In older women we remove a more generous zone of normal breast tissue, and in two instances we encountered a small area of carcinoma adjacent to the clinically obvious fibroadenoma. In this series there were three lesions simulating large fibroadenomas which were histologically demonstrated to be fibrosarcomas in two and cystosarcoma phyllodes in one. Hodgkin's disease was encountered in one patient.

Inflammatory Lesions.—Inflammatory lesions of the breast, such as infected cysts, fat necrosis, plasma cell mastitis, breast abscess, and tuberculosis, may simulate cancer. Small localized inflammatory lesions are treated as breast tumors, with wide excisional biopsy. When the inflammatory lesion is fluctuant with frank pus, the area is incised; a piece of tissue is removed for biopsy; a culture is taken, and a drain is inserted if the frozen section report is benign. When there is diffuse inflammation with induration and cellulitis of the skin, the question of inflammatory carcinoma arises. These patients are given intensive antibiotic therapy while roentgenologic examination is carried out for evidence of distant metastases. Within a few days to a week, if marked improvement has not occurred, a specimen of the breast is taken for biopsy for evidence of cancer. If localization has occurred, the abscess is managed as already described. We had 13 patients who presented this diagnostic problem, and 7 of these were proved to have carcinoma. Two of our patients had tuberculosis of the breast. One of these patients previously had cancer of one breast, and subsequently tuberculosis developed in the other.

Paget's Disease.—Eczematoid lesions of the nipple or areola should be considered malignant until proved otherwise. In some cases of Paget's disease there may be no clinically detectable mass beneath the nipple or in the breast, although the lesion, even at this stage, is cancer. Of our three cases of Paget's disease, one patient lacked a palpable mass. When a mass is palpable, it should be subjected to wide excisional biopsy. When a mass is lacking, a very large core of breast tissue should be removed with the major duct system, and in some instances a simple mastectomy is justified to allow complete search of the breast by the pathologist.

Bilateral Biopsy.—It is important to note that lesions may be multiple in the same breast (9% by pathologic report of our series) or bilateral (26% of this series). In the latter group biopsy was performed on the two breasts, and in eight cases (3.1 %) cancer was found in the less supicious breast. This experience reemphasizes the elusiveness of cancer from the clinical diagnostic standpoint. It also stresses the importance of contralateral breast biopsy when bilateral disease is present and when the frozen section report from the first side is benign. It may be a serious error to assume that cancer is absent in the opposite breast when an apparently similar lesion is reported as benign on the one side.

Biopsy of Recurrent Breast Lesions.—Once surgery has been performed, it is still mandatory that the patient be subjected to periodic examination of the breasts by her physician. If another area develops in either one that varies in consistency from the surrounding tissue, biopsy must again be performed without delay. In

our series, 213 patients (21.3% of our cases) had had previous surgery performed on one or two breasts (Table 6). Of 186 patients who had had benign lesions removed previously, 16 (8.6%) were found to have cancer. Of our 260 cases of breast cancer, 9 patients (3.5%) had had cancer previously in the other breast. It is also interesting to note that the ratio of benign to malignant lesions in the first and second breast biopsies was almost identical. For a more complete analysis of recurrent breast lesions see Table 6 and 7.

Biopsy in Men.—The indications for breast biopsy in men are similar to those in women. In our series 2 (3.8%) of the 53 male patients had cancer (0.8% of the total cancer cases). Since breast tissue is scant in the male and since it can be entirely removed without difficulty or mutilation, our biopsy includes all of the breast tissue plus the subjacent pectoral sheath. We have satisfactorily

TABLE 6.—*Recurrent Breast Biopsy (213 Cases) and Type and Location of Lesion**

Type of Lesion	Same Breast	Opposite Breast	Total	
			No.	%
Present lesion benign	71	114	185	87.0
Present lesion malignant	14	14	28	13.0
Previous lesion benign	79	107	186	87.5
Previous lesion malignant	6	21	27	12.5

* Three biopsies were performed on specimens of each of eight patients during separate hospital admissions, all of which were benign.

TABLE 7.—*Recurrent Breast Biopsy: Analysis of Past Versus Present Biopsies*

	Present Lesion Benign (185 Cases)				Present Lesion Malignant (28 Cases)			
	Same Breast	Opposite Breast	Total		Same Breast	Opposite Breast	Total	
			No.	%			No.	%
Previous lesion benign (186 cases)	68	102	170	91.4	11	5	16	8.6
Previous lesion malignant (27 cases)	3*	12	15	55.5	3*	9	12	44.6

* Recurrent nodule in previous radical mastectomy area.

employed either a transverse incision that skirts the areola or a lateral incision along the anterior axillary fold. In most cases for cosmetic reasons the nipple and areola are not removed. A frozen section is obtained, and, if cancer is found, an immediate radical mastectomy is performed.

OPERATIVE MANAGEMENT

Surgeon-Pathologist Cooperation.—We wish to emphasize that, prior to definitive surgery for any breast lesion, a biopsy should be performed and the frozen section report obtained. Reliance upon seemingly obvious clinical and gross surgical observations cannot safely supplant histologic examination, even in the hands of the most experienced surgeon. The closest coordination between surgeon and pathologist is thus necessary.

Frozen section examination in our institution is not limited to a single section taken only from the most supicious area. Rather, the entire specimen is examined, and all areas supicious for cancer are processed for immediate frozen section, as

well as for later routine paraffin section. On occasion an actual conference between surgeon and pathologist or the insertion by the surgeon of one or more identification sutures to suspicious areas (Fig. 2) may lessen the chance of an area of early malignancy being overlooked.

In 5% of our cancer cases the pathologist was unable to make the diagnosis with certainty on multiple frozen section examinations. In these instances, radical mastectomy was delayed until the histologic proof was obtained three or more days later. We have followed this policy, since in numerous other instances the paraffin sections disclosed benign lesions.

Preparation of Patient.—Breast biopsy must be carefully planned, since it is commonly a preliminary to radical mastectomy. It is essential that the patient be hospitalized, surveyed for metastatic cancer, and evaluated for the risk of a general anesthesia and possible radical mastectomy. It is our custom to have blood available before the biopsy is undertaken. The patient should be fully aware of the need for immediate radical mastectomy if the frozen section report is malignant. In our cases, prior to biopsy, written permission is obtained for any operation deemed necessary by the surgeon, including radical mastectomy. While this may impose a temporary emotional disturbance, it more than compensates for the shock that would follow an unexpected radical mastectomy.

We do not perform breast biopsy on an out-patient basis for the following reasons: (1) The patient is denied the benefit of immediate radical mastectomy should malignancy be encountered; (2) the surgeon must assume that the lesion is benign, and this assumption is highly vulnerable; (3) it encourages the use of local anesthesia, which we consider contraindicated for reasons to follow.

Anesthesia.—All operations in this series were performed with the patients under some form of general anesthesia. Thiopental (Pentothal) sodium was usually employed for short procedures. Ether or some other general inhalation anesthetic agent was employed whenever radical mastectomy was anticipated preoperatively. When a surprise report of a malignant lesion was obtained in a patient under thiopental sodium anesthesia, the anesthesia was switched to ether. We do not use local anesthesia for the following reasons: (1) It tempts the surgeon to restrict the size of the biopsy; (2) it tempts the surgeon to perform the procedure on an out-patient basis; (3) it imposes a poorly timed psychologic shock upon a fully consious patient if she must submit to general anesthesia for immediate radical mastectomy with the sudden full realiziation that she has cancer. The temptation to use local anesthesia is greatest in patients with apparently benign lesions and in whom the psychologic shock would be greatest should a malignant lesion be encountered.

Surgical Technique.—Prior to surgery the tumor area is outlined on the overlying skin with indelible ink.[2] If additional suspicious areas exist in either breast, they are similarly marked. The operative field is prepared widely with iodine and alcohol.

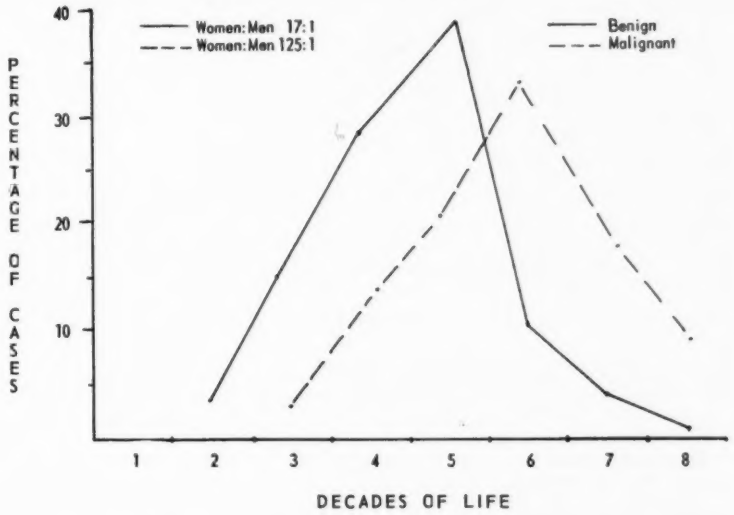
In our cases a liberal radial incision was used overlying the tumor. In many instances, particularly those in which cancer was suspected in association with cystic hyperplasia or duct papilloma, the incision extended from the outer margin of the areola to the periphery of the breast. We have long abandoned the use of curvilinear incisions at the periphery of the breast and "peep-hole" incisions which cater to cosmetic results, often at the expense of adequate exposure or size of the specimen for biopsy.

2. Shallow, T. A.; Eger, S. A., and Wagner, F. B., Jr.: Scientific Exhibit: Management of Breast Lesions, Postgrad. Med. **8**:499-505, 1950.

As already stated, the area of excision varies with the nature and extent of the lesion. In all instances the local lesion is excised with a wide margin of surrounding tissue. When cancer is being sought in older women who have had long-standing cystic hyperplasia, the area of removal is much wider and may include one or more entire quadrants. After the core of breast tissue has been removed, the breast is carefully palpated through the incision for other areas suspicious of being malignant. Any such areas encountered are also removed and sent to the laboratory for immediate frozen section examination. If no cancer is found, reconstruction of the breast is undertaken to minimize postoperative deformity. This is accomplished by the proper placement of sutures to obliterate dead space and to restore the breast to a normal contour.

The patient is kept in the operating room and under light anesthesia until the report of the frozen section is received. This avoids any unnecessary prolongation of the procedure, confusion, or embarrassment should a report of a malignant lesion be obtained. Neither the

FIGURE 3.—Age Incidence of Benign vs. Malignant Breast Tumors



patient nor the family is given a final opinion regarding the nature of the breast lesion until the paraffin sections have been examined histologically, since, as previously stated, in 5% of our cancer cases the pathologist was unable to make the diagnosis with certainty on frozen section examination.

ANALYSIS OF RESULTS

Although the cancer group in this series was only 260 cases (26%, Table 1), each patient was considered a cancer suspect, whether remote or real. The clinical evaluation, criteria for biopsy, and operative technique were uniformly employed, since the patients were all personally managed by the senior author. The cancer cases included in this series were all operable.

Figure 1 shows the well-established fact that the incidence of breast cancer rises sharply with increasing age. Figure 3 portrays the curves for age incidence of benign and malignant tumors. It is first seen that the patients for all types of lesions predominate in the fourth to sixth decades of life. Next, the age incidence

for benign lesions precedes and parallels that for the malignant ones by a decade. Finally, the drop in incidence of malignant lesions after the sixth decade is due to the smaller number of people living in that age group, although the relative incidence of cancer in this group, as shown in Figure 1, is much higher. For benign lesions women predominate 17 to 1; whereas for malignant lesions they predominate 125 to 1.

Figure 4 demonstrates that the average duration of symptoms until biopsy for patients with either benign or malignant lesions was approximately the same. This

FIGURE 4.—Duration of Symptoms of Benign vs. Malignant Breast Tumors

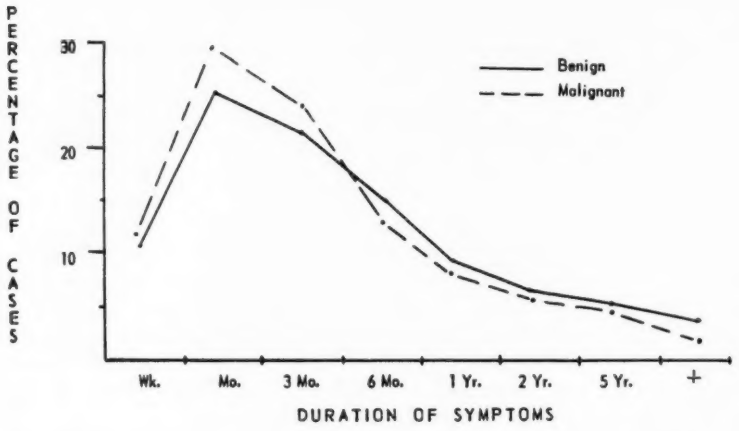


TABLE 8.—*Comparative Distribution of Benign and Malignant Lesions in the Breast*

Location	Benign, %	Malignant, %
Upper outer quadrant (including axillary tail of Spence)	51.8	48.9
Lower outer quadrant	8.0	13.3
Upper inner quadrant	7.0	10.4
Lower inner quadrant	5.8	7.9
Beneath the nipple and/or areola	9.7	12.1
Mass near the areola	5.9	8.3
Diffuse involvement	14.2	6.2
Shotty	9.7	3.1
Multiple nodules	4.5	3.1
Nothing palpable in the breast (pain or discharge present)	3.5	1.2

would seem to indicate that the reaction of the patient, as well as the examining physician, is the same for any breast lesion. Although the majority of patients underwent biopsy within three months of onset of symptoms, regrettably over one-fifth delayed a year or more before biopsy.

The location of benign and malignant lesions in the breast is analyzed in Table 8. There was no significant preponderance between right and left sides, and for this reason the figures given are approximately for either breast. It will be noted that the predominant site for all breast lesions is the upper outer quadrant (approximately 50%). The remaining quadrants are roughly equally involved, with possibly the lower inner quadrant being the least frequent site. In this series

the nearness of the lesion to the nipple or areola did not appear particularly to indicate a malignant lesion. It is of great interest to note from Table 8 that in 6.2% of the malignant lesions there was only diffuse change in the breast without a single mass, and in 1.2% of the malignant lesions there was no palpable change in the breast, with biopsy indicated only because of pain or discharge.

Table 2 shows the relationship of tumor size to malignancy. While larger tumors were more apt to be malignant, we wish to stress that the smallest lesion can be malignant and that the size of the tumor should not be relied upon as a diagnostic criterion. In this series approximately 20% of the malignant lesions were less than 1 cm. in diameter.

Table 3 shows the lesions found associated with cancer. In a great number of these cases the benign lesion was suspected clinically, and the malignant lesion was discovered incidentally at the time of biopsy.

The clinical features of the lesions in this series are analyzed in Table 9. Approximately 75% of the patients with malignant tumors had the characteristic single

TABLE 9.—*Clinical Features*

	Benign		Malignant	
	No.	%	No.	%
Single mass	348	47.0	207	76.5
Shotty area(s)	239	32.3	26	10.0
Thickened area	94	14.7	13	5.0
Multiple nodules	49	6.6	8	3.1
Discharge from nipple	49	6.6	11	4.2
Nothing palpable (pain, tenderness, or discharge present)	30	3.5	3	1.2
Rapid growth of lesion	8	1.1	24	9.2
Skin fixation	10	1.4	96	36.9
Axillary nodes involved				
Clinically	19	2.6	70	26.9
Pathologically (cancer)	..	....	124	48.6
Pain	178	24.1	36	13.9
Tenderness	160	21.6	32	12.3
Pain and/or tenderness	263	35.6	58	22.4

mass. The remaining 25% presented delusive findings, such as one or more shotty areas (10%), only a thickened area (5%), multiple nodules (3.1%), discharge from the nipple (4.2%), or even nothing palpable in the breast (1.2%). The presence of pain or tenderness should not militate against the diagnosis of cancer or unduly favor the diagnosis of cystic hyperplasia, since 13.9% of the cancer patients had pain; 12.3% had tenderness, and 22.4% had either one or the two symptoms. The causes and types of discharge from the nipple are shown in Tables 4 and 5. As previously stated, we regard all patients with any type of discharge from the nipple as cancer suspects, since 18.3% of these patients in our series had cancer.

Tables 6 and 7 present an analysis of recurrent breast lesions. It is noteworthy that the ratio of benign to malignant lesions in the first and second biopsies was approximately the same. This seems to indicate that a recurrent breast tumor has the same chance of being malignant as one seen in a breast for the first time.

SUMMARY AND CONCLUSIONS

Earlier diagnosis of the breast cancer demands prompt biopsy of any localized area that varies from the surrounding tissue. "Watchful waiting" is a dangerous policy that may deprive the patient of treatment during the period of curability.

Adequate biopsy consists of the removal of sufficient breast tissue to reveal the presence of any obvious or occult cancer by immediate thorough pathologic examination. Cancer was found in 26% of 1,000 consecutive patients operated on, the following criteria being used:

1. Wide excisional biopsy is particularly indicated when the clinical diagnosis is thought to be localized cystic hyperplasia, since unsuspected cancer may be found or may coexist.

2. All patients with discharge from the nipple are cancer suspects, whether a mass is palpable in the breast or not. A wide excisional specimen for biopsy is indicated, since multiple lesions are frequently present.

3. One should not hesitate to perform bilateral breast biopsy, even when the frozen section report from the more suspicious side is benign.

4. Recurrent tumors of the breast deserve biopsy just as promptly as the initial ones, since the chances of cancer are just as great.

5. Biopsy of the male breast should not be neglected, since cancer does occur. We advocate removal of all breast tissue for examination, with preservation of the nipple.

Proper management includes (*a*) preoperative study to rule out metastasis or surgical handicaps; (*b*) psychologic conditioning of the patient to the possibility of radical mastectomy; (*c*) the availability of blood; (*d*) close liason between surgeon and pathologist; (*e*) avoidance of local anesthesia and out-patient biopsy; (*f*) operating room and personnel available for immediate radical mastectomy, if necessary.

There are no clinical signs that reassure against cancer. Cancer was sometimes found in patients whose presenting diagnostic criteria were in favor of benign lesions (13.5% of cases in this series).

Pain or tenderness in the breast was present in 22.4% of the cancer patients.

The smallest lesion may well be malignant, since 20% of the cancer patients had lesions 1 cm. or less in diameter.

Patients with discharge from the nipple had cancer in 18.3% of cases.

Cancer was associated histologically with cystic hyperplasia in 15.4% of cases. Clinically, many of these cases were not suspected.

In 6.2% of the cancer cases there was a diffuse, rather than localized, change in the breast.

Cancer existed in the less suspicious breast in 3.1% of our patients subjected to bilateral biopsy. A benign report from the first side should not dissuade the operator from performing biopsy on the other side.

LAROQUE APPROACH TO INGUINAL HERNIAS IN PEDIATRIC PATIENTS

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THE EARLY repair of inguinal hernias in pediatric patients has not been universally accepted as the treatment of choice, despite an increasing number of articles in the literature recommending early operation.[1] It is the purpose of this paper to substantiate the idea of early repair based on our experience with 45 cases in the past three years and particularly to advocate LaRoque's intra-abdominal approach in the repair of these herniae.

For practical purposes, all inguinal hernias in patients in this age group are of the indirect type. The pathology of the indirect hernia consists of two deviations from the normal.[2] First, there is a preformed sac of congenital origin, when the processus vaginalis fails to obliterate, retaining an opening with the peritoneal cavity. These hernias are not due to muscle or fascial weakness of the abdominal wall. In the absence of a persistent processus vaginalis, indirect hernia does not develop irrespective of the force or violence applied to the abdomen. Secondly, when the bowel or omentum enters the sac, the internal ring becomes secondarily dilated. Consequently, to effect a cure of this type of hernia all that is required is simple excision of the sac and closure of the internal ring to its normal circumference, if it is dilated.

Conservative treatment is often recommended by pediatricians and surgeons alike. In this age group, if the child is otherwise healthy, it is our belief that surgical repair of these herniae should be performed when recognized. If conservative treatment is followed, the hernia enlarges and becomes more difficult to repair. The child's activities are limited; a cumbersome truss is frequently worn; incarceration and strangulation occur with a surprisingly high incidence, and adhesions form about the sac rendering dissection more difficult.

From 1949 to 1952 we have operated upon 45 patients between the ages of 5 weeks and 8 years. In this group two patients had bilateral hernias, making a

From the Clinch Valley Clinic.

1. Cogswell, H. D., and Czerny, E. W.: Treatment of Hernias in Infants and Young Children, Am. Surgeon **19**:87-89 (Jan.) 1953. Cresson, S. L.: Management of Hernias in Infants and Children, M. Clin. North America **36**:1767-1777 (Nov.) 1952.

2. Zimmerman, L. M.: Recent Advances in Surgery of Inguinal Hernia, S. Clin. North America **32**:135-153 (Feb.) 1952.

total of 47 herniorrhaphies. The average age of the patients was 2.8 years. Eight patients were 2 months old or younger at the time of operation. There was only one girl in this age group. There were eight patients with incarcerated hernia, indicating the high incidence of this complication in this age group. Twelve associated hydroceles were repaired with the herniorrhaphy. The average postoperative hospital stay was four days. There was no operative mortality. No wound infection or other morbidity occurred. Follow-up in these cases to date has revealed no recurrence. The intra-abdominal approach of LaRoque was utilized in 37 instances.

The difficult part of the operative repair of these hernias is the identification and dissection of the sac. Most authors agree that simple excision of the sac is all that is necessary to effect a cure. In these patients, tissues are delicate and the sac frequently small and thin walled. When the conventional approach is used, considerable dissection is often necessary to isolate the sac. This dissection produces unnecessary trauma to the cord. As emphasized by Williams in 1938,[3] we have found LaRoque's intra-abdominal approach particularly helpful in identifying the sac in these small patients. Only a few surgeons utilize the intra-abdominal approach in the repair of these hernias, and this procedure has not received the wide acceptance it deserves, despite a number of reports in the literature pointing out the advantages of this technique.[4] It is not mentioned in Watson's textbook on hernias or in Ladd and Gross's textbook on abdominal surgery of infancy and childhood.[5]

LaRoque first described his technique in 1932.[4a] It is as follows: The usual inguinal incision is made and dissection carried down through the fascia of the external oblique muscle. The cord is not elevated. The fibers of the internal oblique and transversalis muscles are separated above the internal ring and the peritoneum opened as in a McBurney incision. The neck of the sac with its thickened peritoneum is readily visualized and the abdominal contents of the sac, if any, are easily evacuated under direct vision. The neck of the sac is then severed from the peritoneum, thus accomplishing complete exclusion of the sac from the abdominal peritoneal cavity, not merely closure of the aperture through which the hernia passed. The likelihood of redundant peritoneum and accessory sacs is thus obviated. The peritoneum is closed transversely. The internal oblique and transversalis muscles are approximated with interrupted sutures.

We have routinely excised the sac because of the possibility of hydrocele formation, although many authors claim this is unnecessary, stating that the sac obliterates when disconnected from the abdominal cavity. With the neck of the

3. Williams, C.: The Advantages of the Abdominal Approach to Inguinal Hernia, Ann. Surg. **107**:917-922 (June) 1938.

4. (*a*) LaRoque, G. P.: The Intra-Abdominal Method of Removing Inguinal and Femoral Hernia, Arch. Surg. **24**:189-203 (Feb.) 1932. (*b*) Jacobson, P.: Inguinal Herniorrhaphy from the Intra-Abdominal Perspective: Retention of Sac and Intact External Ring, and Means for Proving Adequacy of Repair, Am. J. Surg. **71**:797-808 (June) 1946. (*c*) Jacobson, P.: Intra-Abdominal Approach to Inguinal Herniorrhaphy, ibid. **79**:557-568 (April) 1950. (*d*) Williams.[3]

5. Ladd, W. E., and Gross, R. C.: Abdominal Surgery of Infancy and Childhood, Philadelphia, W. B. Saunders Company, 1941. Watson, L. F.: Hernia: Anatomy, Etiology, Symptoms, Diagnosis, Differential Diagnosis, Prognosis, and Treatment, Ed. 3, St. Louis, C. V. Mosby Company, 1948. Williams.[8]

sac in hemostats, the finger is inserted into the sac and dissection from the cord is easily accomplished with minimal trauma to the cord structures. No repair of the inguinal canal is necessary. Rarely, one encounters a widely dilated internal ring, and, when this occurs, simple reconstructive repair of the ring is performed, the transversalis fascia being sutured to the inguinal ligament. If there is an associated hydrocele, this is easily repaired through the same incision and the excess sac excised. The fascia of the external oblique is approximated with interrupted sutures of fine cotton, and clips are applied to the skin.

SUMMARY AND CONCLUSIONS

Inguinal herniorrhaphy is a safe procedure in pediatric patients and deserves wider acceptance.

The intra-abdominal approach of LaRoque gives the surgeon a distinct advantage in the repair of these hernias.

From 1949 to 1952 there were 47 inguinal herniorrhaphies performed upon 45 pediatric patients from 5 weeks to 8 years of age. No mortality occurred, and no recurrence has been observed.

EXPERIMENTAL SUBPHRENIC ABSCESS

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A COMPLETE review of the literature revealed no reported experimental method for the production of subphrenic abscesses. A predictable standardized method would be valuable in studying the treatment and natural history of such a condition. It would be a great aid in the experimental evaluation of the role of surgery, in conjunction with the antibiotic and chemotherapeutic agents, in the treatment of subphrenic abscesses. It was with these aims in mind that the following study was carried out.

MATERIALS AND METHODS

The abscesses were produced in healthy, adult, male rabbits. The basic principle utilized was the fact that a gauze sponge, when placed in the peritoneal cavity of animals or of human beings produces a granulomatous reaction, with the sponge encapsulated in its center. It was felt that the most logical procedure would be to insert a gauze sponge in the subdiaphragmatic space at one operation, allow time for the production of the granulomatous reaction, then reoperate and inoculate the granuloma with a pure culture of bacteria.

The Curity [1] radiopaque sponges were used. These sponges contain a rectangular segment of cloth which has been impregnated with barium sulfite. A segment of sponge containing the marker was used so that x-rays might be taken postoperatively. This would serve as a check on the position of the sponge and on the development of air in the abscesses.

Three series of rabbits were used. One series (Group A) consisted of rabbits operated upon one time, simply for sponge insertion. The second series (Group B) consisted of rabbits on whom a one-stage procedure was attempted; that is, a marked sponge, already inoculated with organisms, was inserted at the first and only operation. A third series (Group C) consisted of rabbits subjected to the two-stage procedure described above.

The organism used in this experiment was Escherichia coli. One cubic centimeter of a 24-hour-old, glucose broth, subculture of a virulent E. coli, isolated from a known, pure clinical abscess (perirectal), was used as the inoculum in each experiment.

The rabbits used were all healthy adult male rabbits varying in weight from 4 lb. to 7 lb.-10 oz. (1.8 to 3.4 kg.). These rabbits were starved for 24 hours preoperatively because of the death of one rabbit from aspiration of food with resultant atelectasis and respiratory obstruction. The anesthesia used was pentobarbital (Nembutal) sodium given intravenously. For each 5 lb. (2.3 kg.) of weight, 60 mg. of pentobarbital sodium was given intravenously (via ear vein). The operating time was usually from 20 to 30 minutes for each of the two stages.

The first-stage operation was performed through a subcostal or transverse incision. Sterile precautions and technique were used throughout. If a second-stage procedure was carried out, the old incision was usually reentered. However, some of the second-stage operations were performed through a longitudinal muscle-splitting incision. At first, cotton was used as suture

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1. These sponges are manufactured by Bauer & Black, Division of the Kendall Company, Chicago.

material. However, the development of several subcutaneous abscesses about cotton sutures led to a change to steel wire. The sponge was made into a 2 cm. ball (with the radiopaque marker in its center) by means of a 000 cotton purse-string suture, and attached to the diaphragm with a 000 cotton suture in such a way as not to penetrate the thoracic cavity with the needle. The site chosen was over the right anterior lobule of the rabbit liver.

The rabbits were fed regular animal house diets and were weighed once every 7 to 10 days. Since the temperatures of the rabbits varied with the temperature of the room and with the excite-

Protocols

Group A.—One operation for insertion of sponge

RABBIT 14.—This animal was killed five months postoperatively. The sponge was well encapsulated, forming a 1 cm.-sized mass, with a wall measuring 2 mm. in thickness. This mass was between the liver and the diaphragm, with the omentum adherent anteriorly.
RABBIT 15.—The animal died an operative death, with atelectasis.
RABBIT 18.—This animal was killed 19 months postoperatively. The sponge was walled off within a 1.5 cm.-sized mass with a wall measuring 1 mm. in thickness. Adherent to this mass were stomach, diaphragm, and liver.
RABBIT 19.—This animal died an operative death, with massive atelectasis following aspiration.
RABBIT 20.—This animal was discarded in error. No mass was palpable at the time of death, 10.5 months after operation.

Group B.—One operation for insertion of sponge already inoculated with E. coli

RABBIT 21.—Four and one-half months after operation this animal was killed. There was a mass measuring 1 cm. in diameter, between the liver and diaphragm. Adherent to this mass were the kidney, the omentum, the liver, and the diaphragm. The wall of the mass was 1 mm. thick and enclosed the sponge.
RABBIT 22.—This animal was killed five months after operation. There was a 1.5 cm. diameter mass with a 2 mm. thick wall surrounding the sponge. The liver, diaphragm, and omentum were adherent to the mass. Aspiration culture of the mass was sterile.
RABBIT 23.—This animal died an operative death.
RABBIT 24.—This animal was discarded in error. No mass was palpable at the time, nine months after operation.
RABBIT 25.—This animal died an operative death, with pneumothorax and atelectasis.
RABBIT 26.—This animal died an operative death. The cause was undetermined.

Group C.—Two-stage operation

RABBIT 10.—Death occurred four months after the first operation, and 3.5 months after the second. There was a mass 7 by 5 by 4 cm. occupying the right upper quadrant. This mass had a 3 mm. thick wall and contained yellow purulent material surrounding a sponge. The culture yielded E. coli. Adherent to this mass were the diaphragm, liver, colon, ileum, and anterior abdominal wall. There was also a generalized peritonitis with a moderate ileus. Culture of this also yielded E. coli.
RABBIT 11.—This animal was killed 6.75 months after the first operation, and 6 months after the second. There was a mass 9 by 7 by 6 cm. occupying the entire right upper quadrant. The mass had a 3 to 5 mm. thick wall and contained yellow purulent material surrounding a sponge which was still sutured in place. Culture yielded E. coli. Adherent to and displaced by this mass were the diaphragm, liver, colon, ileum, omentum, and anterior abdominal wall (Figs. 3 and 4).
RABBIT 16.—This animal died immediately after the second operation, which was performed three weeks after the first. Autopsy revealed a mass 2 by 1 by 1 cm., consisting of a sponge walled off between the liver and the diaphragm, with omentum and colon adherent anteriorly. Culture of the mass was sterile. There were no other abnormalities.
RABBIT 17.—The findings were the same as those listed for Rabbit 16.
RABBIT 27.—This animal died immediately after the second operation, four weeks after the first procedure. The findings were the same as those listed for Rabbit 16.
RABBIT 28.—The findings were the same as those listed for Rabbit 27.
RABBIT 29.—This animal was killed 15 months after first, or 13.5 months after the second, operative procedure. There was a 4 by 4 by 3 cm. mass with a 2 mm. thick wall. This mass contained some thick, white, cheese-like material and the sponge. The liver, diaphragm, omentum, stomach, and duodenum were adherent to the mass.
RABBIT 30.—This animal died 15 months after the second, and 16.5 months after the first, operative procedure. There was a mass 4 by 5 by 1 cm. presenting between the diaphragm and the liver. The mass had a wall measuring 2 mm. in thickness and contained yellow purulent material and the implanted sponge. The liver, diaphragm, pylorus, gall bladder, stomach, colon, and ileum were adherent to the mass. Patchy atelectasis and pneumonia were also present.
RABBIT 31.—This animal died immediately after the second, and 3.25 months after the first, procedure. Autopsy revealed a mass 3 by 2 by 1.5 cm. with a 2 mm. thick wall containing the implanted sponge. The pylorus, omentum, duodenum, liver, and diaphragm were adherent to the mass. Bronchitis and pneumonia were present.
RABBIT 32.—This animal was discarded in error approximately seven months after the second operative procedure, with a palpable abdominal mass present.

ment of the animals, the use of temperature readings as indicators was discontinued. X-rays were taken every two to four weeks. Both anteroposterior and right lateral erect films were taken each time.

Physical examinations of the rabbits for the presence of abdominal masses were carried out in the supine and in the prone standing position. It was found possible to feel an abdominal mass more easily in the latter position.

RESULTS

Group A.—This group consisted of five rabbits which served as controls. Each of these was subjected to a one-stage operation for the implantation of a sterile sponge. One of these animals died during the operation. One died postoperatively

of aspiration of food with resulting respiratory obstruction. The postoperative course of the other rabbits was uneventful. For approximately two months a mass was palpable in the subcostal region when the rabbit was placed in the prone position. This mass was the size of the implanted sponge and never became larger. It actually receded beneath the costal margin after this time and subsequently could not be palpated. The rabbits remained healthy, ate well, and gained weight. Roentgenograms of these rabbits revealed that the sponges remained where they were implanted. In two of these rabbits there appeared to be a rim of air surrounding the sponges (Figs. 1 and 2).

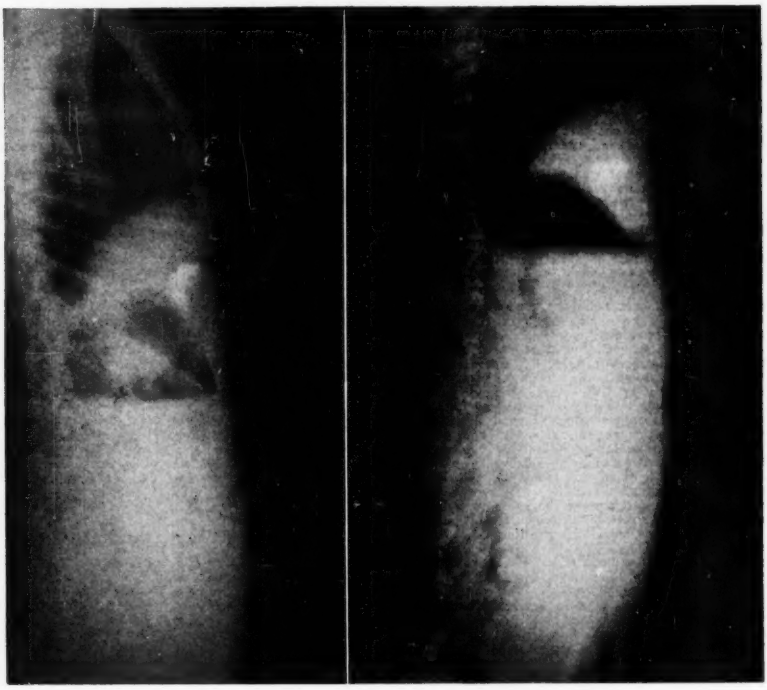

Fig. 1.—X-rays showing radiopaque marker beginning to uncoil, with a narrow rim of air about periphery of marker. Small areas resembling calcification are seen. Steel suture lines are visible.

An idea of what happened to these sponges was obtained from the findings at the time of the second-stage operations done on the rabbits in Group C and from the autopsies of the Group A animals. The second operation was done two weeks after the first procedure in one rabbit, three weeks after in three of the rabbits, four weeks after in three of the rabbits, five weeks after in two of the rabbits, and seven weeks after in one rabbit. Autopsies were done on Group A animals 5 and 19 months postoperatively. Two weeks after the insertion of a sterile sponge there was found a fibrinous peritonitis above the entire right anterior lobule of the liver, between the diaphragm and the superior surface of the liver. Overlying the right anterior lobule

of the liver there was a mass surrounded by fibrous pinkish-white adhesions adherent to both diaphragm and liver, with the sponge in the center of the mass, and surrounded by sterile (proved by aspiration and culture) white fluid. At the end of three weeks the same conditions prevailed, except that the process was intensified and a distinct wall had formed. In addition, the omentum became adherent to the mass and to the liver, anteriorly. After four weeks the adhesions were more fibrous and the omentum, colon, and stomach pulled up to the mass anteriorly. The mass still was adherent to the liver and the diaphragm. There was no change in the situation after five or seven weeks. Autopsy 5 and 19 months after the original operation revealed the sponge to be in place, surrounded only by a thin, translucent, fibrous, veil-like wall. The right lobe of the liver was partially adherent to the sponge

Fig. 2.—X-rays showing presence of a mass, in center of which there is a radiopaque marker. Rim of air can be seen about sponge marker. Mass is pushing diaphragm cephalad and gas-containing viscera caudad and laterally.

and these, in turn, to the diaphragm. The omentum was adherent to the liver edge and to the sponge. The stomach and colon were in normal position. There was no gross evidence of abscess formation or peritonitis. The fifth rabbit in Group A died about 10 months postoperatively and was discarded by the animal house caretaker. No autopsy was performed.

Group B.—This group consisted of six rabbits subjected to a single-stage operation. The operation was the same as that done in Group A, except that the sponge used had already been inoculated with 1 cc. of the E. coli culture. One rabbit was found dead the next morning and was proved to have a pneumothorax with atelectasis of the entire right lung. A second rabbit died postoperatively but was discarded by the animal house caretaker before an examination could be made. A third

rabbit died immediately postoperatively, and no cause of death was found at autopsy. The postoperative course of the remaining three rabbits was essentially uneventful and similar to that of the Group A rabbits. Autopsy and roentgenographic findings in this group were the same as those in the Group A rabbits.

Group C.—This group was composed of 10 male rabbits subjected to a two-stage operation. The first operation was the same in all of the rabbits. The course of events has been described under the Group A rabbits. The second operation was carried out at intervals ranging from two to seven weeks after the first stage. The abdomen was reopened and the mass located and allowed to remain as undisturbed as possible. An aspiration culture was taken. (All aspiration cultures taken at the second operation were negative for pathogens.) One cubic centimeter of the E. coli inoculum broth was injected into the center of the sponge by means of a syringe and a No. 25 gauge hypodermic needle. None of the inoculum was spilled into the wound. Gloves and drape towels were changed after the inoculation in order to be completely certain of having no accidental contamination of the peritoneal cavity itself or of the wound. Closure was carried out with No. 32 stainless-steel wire sutures.

There were seven deaths following the second-stage operation. Two of the animals died during the night after operation. Autopsies revealed no explanation for the deaths. Three rabbits never awakened from the anesthetized state. The sixth death was that of a rabbit in which a large subdiaphragmatic abscess developed, but the rabbit died suddenly 5 weeks after the second-stage operation, and 13 weeks after the first operation. The seventh death was that of one of the rabbits in which a large subdiaphragmatic abscess also developed. This rabbit gradually went downhill, losing weight and appetite, and a large intra-abdominal mass developed. This rabbit died three months after the second operation, and three and one-half months after the first.

An eighth rabbit in this series was killed six months after the second-stage operation. In this rabbit a 9 by 7 by 6 cm.-sized mass developed in the upper abdomen, but the animal had remained in good health.

The ninth death took place seven months after the second operative procedure, and eight months after the first. A large mass had developed also in this rabbit. Unfortunately, however, this animal was discarded before autopsy could be performed. In the 10th rabbit a subdiaphragmatic abscess developed, and this animal was killed 13.5 months after the second operation, and 14 months after the first procedure.

The sequence of events in all of these rabbits was almost the same. Two weeks after the second operation a mass was palpable under the right costal margin. This mass usually became clearly palpable when it measured 2 or 3 cm. in diameter. By the end of two to three months the mass had increased to 4 to 6 cm. in diameter. X-rays confirmed the presence of the marked sponge within the mass. The mass continued to grow until at the end of six months (if it had not perforated into the general abdominal cavity) it was approximately 7 or 8 cm. in diameter. If the mass remained encapsulated, the rabbit continued in excellent health. In one rabbit, which was killed 13.5 months after the second-stage operation, the mass had remained 4 cm. in diameter until 10 months before autopsy.

In the rabbit which was killed after six months, a 9 by 7 by 6 cm.-sized mass had developed in the right upper quadrant. At autopsy (Fig. 3) this mass was found to be yellow and thick walled, with numerous blood vessels coursing over its surface. The mass was bound to the anterior abdominal wall and the diaphragm by fibrous adhesions. The right diaphragm was markedly elevated. The liver was pushed to the left and caudad. The stomach was pushed to the left and compressed. The greater omentum, colon, and two loops of small intestine were drawn up to the mass and adherent in the right upper corner of the right upper quadrant. The mass pressed on the great vessels posteriorly. There was no generalized peritonitis. An

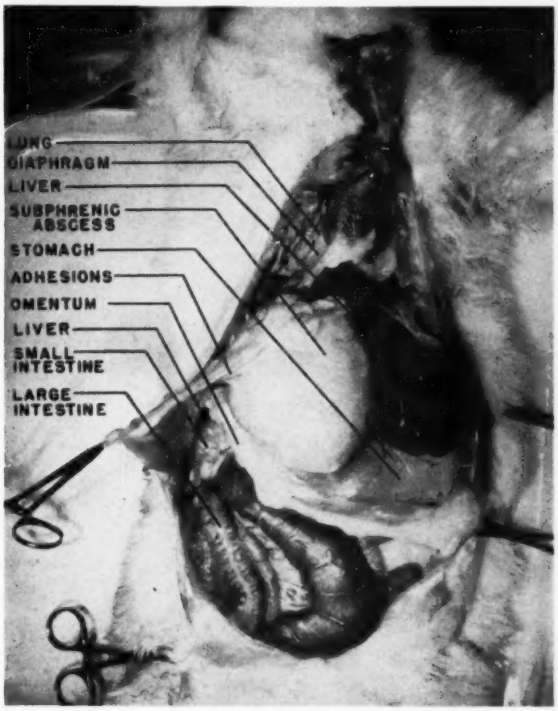

Fig. 3.—Autopsy of rabbit in which subdiaphragmatic abscess developed.

aspiration culture taken under sterile precautions yielded pure E. coli. Incision into the abscess revealed about 150 cc. of thick, yellow, creamy, foul-smelling fluid within the thick-walled sac. In the center of this sac the sponge was located, with the cotton suture still holding it to the diaphragm. The rabbit which was killed after 13.5 months showed a 4 cm.-sized mass, yellowish-white, bound down by fibrous adhesions between the mass, the diaphragm, the liver, the stomach, and the omentum. The mass on section had a 2 mm. thick wall and revealed the presence of a caseous white material surrounding the sponge.

In the seventh rabbit mentioned, the sequence of events fell within the usual range for two months. Then a second smaller mass developed just to the left of the

original mass. The two masses continued to increase in size simultaneously. The mass which occupied mainly the right upper quadrant reached a 7 cm. diameter by palpation. The smaller mass in the epigastrium and under the left costal margin reached a diameter of 3 cm. The rabbit gradually began to lose appetite and weight

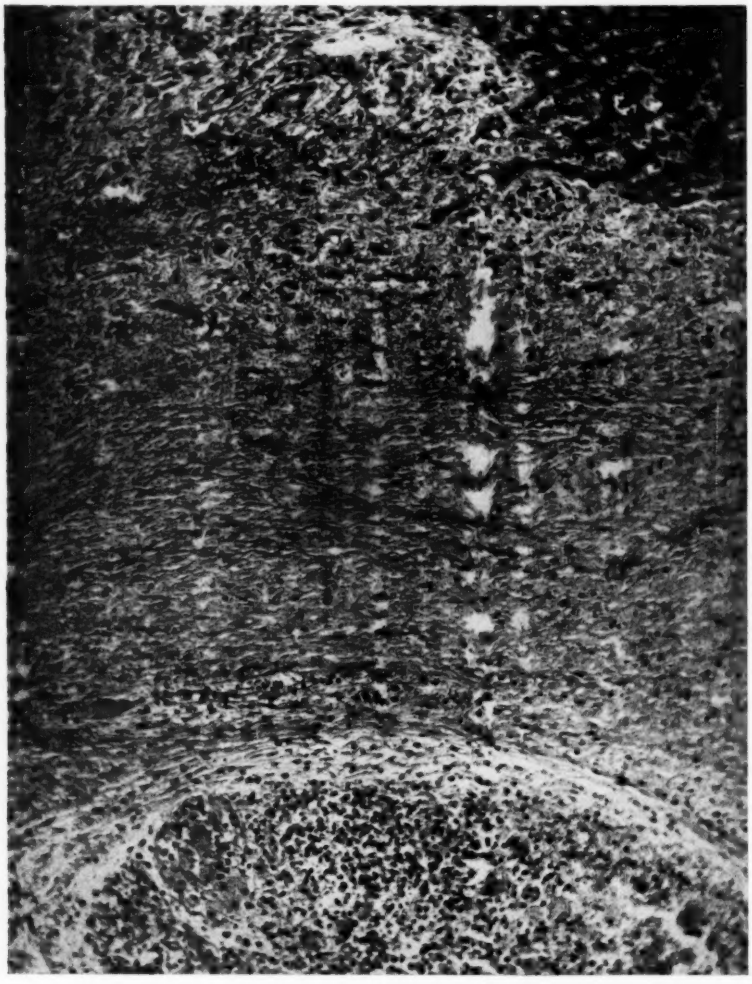

Fig. 4.—Photomicrograph (see text for description of microscopic findings).

and suddenly died, three months after the second-stage operation. Autopsy revealed a 7 by 5 by 4 cm.-sized mass in the right subdiaphragmatic space, with almost the same findings as described previously. However, the mass which had been felt in the left subcostal and epigastric regions was gone. Apparently it had ruptured spontaneously, for there was generalized peritonitis throughout the abdomen, with

a paralytic ileus. There was a yellow fibrinopurulent exudate over the dilated loops of large and small bowel. A culture of the abscess, taken under sterile precautions through the right diaphragm, yielded E. coli, as did a culture of the peritoneal pus. The remainder of the autopsy findings were negative.

MICROSCOPIC FINDINGS

The sections taken at autopsy from the various animals showed a varied picture (Fig. 4). In some there was a minimal degree of fibrous reaction about large intact remnants of gauze sponge, fragments of which were surrounded by numerous

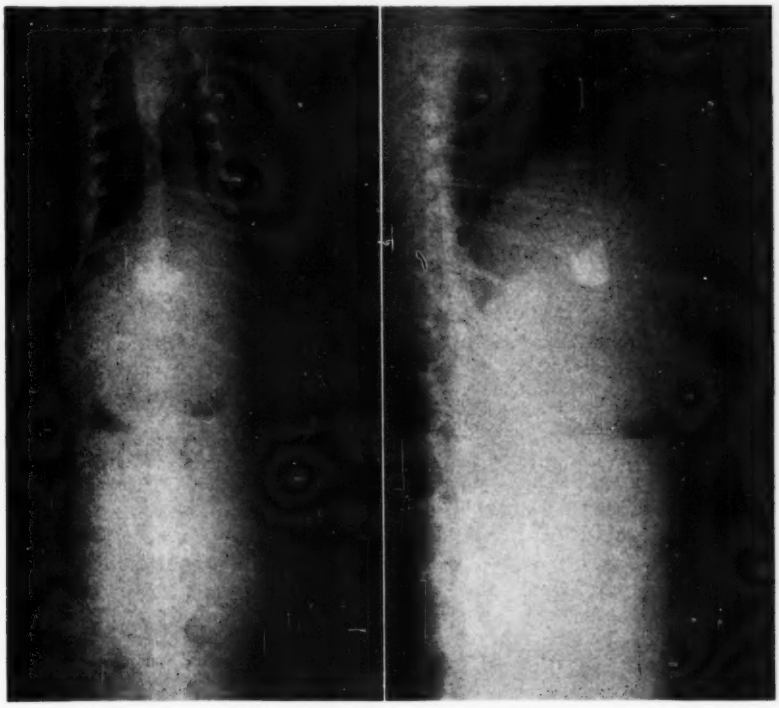

Fig. 5.—Same rabbit as pictured in Figure 2. Note increased size of the mass.

foreign-body giant cells. There was a moderate degree of lymphocytic and polymorphonuclear leucocytic reaction. In other sections there was a strikingly thick fibrous capsule composed of dense collagenous fibrous tissue surrounding a dense collection of polymorphonuclear leucocytes (the abscess cavity).

ROENTGENOGRAPHIC FINDINGS

Roentgenograms were taken on all of the rabbits, beginning two weeks postoperatively. These were repeated every two to four weeks. No air was present about the sponges in the first set of x-rays taken of the Group A series. However, in the three surviving rabbits of this group, a small rim of air was found surrounding

the marked sponge on x-rays taken five months after the first operation. There were also scattered irregular areas of what might have been calcification around the radiopaque marked sponge, and in one rabbit there appeared to be uncoiling of the marker, which, in turn, meant of the sponge (Fig. 1). The marker consisted of gauze impregnated with barium sulfite. Therefore what appeared to resemble calcification on the x-ray might have been deposition of barium salts. In one rabbit a steel-wire marker was substituted for the barium marker without the subsequent appearance of these roentgenographic findings.

In the Group B series the x-rays revealed no air about the sponges, no uncoiling of the sponge, and no deposits resembling calcification.

In the Group C series it was possible to trace the increase in size of the abscess about the sponge (Figs. 2 and 5). The position of the surrounding gas-filled viscera changed with the growth of the mass. The right diaphragm became elevated, with displacement of the heart to the left. In addition, beginning about three months after the inoculations of the sponges, a rim of air became visible about the radiopaque marker. An air-fluid level was never seen.

SUMMARY

The experimental production of a subphrenic abscess is described. The method utilized was a two-stage procedure in which the first operation consisted of the introduction of a foreign body and the development of a granulomatous mass, followed by a second operation for the inoculation of this mass with a known culture of Escherichia coli.

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Dr. Tobias Weinberg, Pathologist-in-Chief and Director of Laboratories, gave valuable advice and was responsible for the interpretation of the microscopic sections.

Valuable assistance was given by the Crockin Laboratory Technicians, Messrs. Jack Circle and Thomas Hoffmaster, who developed an animal holder and cassette combination which aided immeasurably in the taking of x-rays.

COMPLICATIONS ASSOCIATED WITH THE USE OF "CONTROLLED HYPOTENSION" IN ANESTHESIA

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AND
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THE TECHNIQUE of "controlled hypotension," introduced primarily for the prevention of excessive bleeding during surgical operations, has been embraced by an ever-increasing number of anesthetists and surgeons. In most instances, no doubt, the popularity of the technique is based on its inherent value. However, for those who may be attracted to it by reason of its break with traditional concepts or because of the uninhibited enthusiasm of many published reports, it must be emphasized that multiple dangers accompany its use. The proper evaluation of the technique and a true estimate of its position in anesthetic and surgical practice are dependent upon knowledge both of these complications as well as of the beneficial effects possible for the individual patient.

In 1946 Gardner [1] introduced the technique of arteriotomy, whereby blood is withdrawn from the patient preoperatively via the arterial route and then replaced in part or *in toto* by the same manner at the end of operation or during operation itself, if necessary. Such preoperative reduction of the total circulating blood volume serves to lower the blood pressure and at the same time to produce peripheral vasoconstriction [2]; when combined with the proper positioning of the patient, the net result is a reduction of bleeding at the operative site. Arteriotomy approximates a state of hemorrhagic shock and thus exceeds the bounds of physiologic propriety. A more physiological method, introduced by Gillies in 1948, consists of preganglionic total sympathetic blockade.[3] In this method the blood pressure is reduced by the use of spinal anesthesia to achieve paralysis of the entire efferent sympathetic outflow, with slighter degrees of sensory and motor blockade, so that the muscles of respiration and the medullary centers remain relatively unaffected. Gillies has admitted that even this technique is "physiologic trespass" [4] but has assumed that the arteriolar dilatation so produced guarantees normal capillary circulation even in

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the face of a reduced systemic pressure head, provided that the total blood volume is within normal limits and inherent capillary tone remains unaltered. Proper posturing of the patient is again an essential feature for the control of bleeding, as it is, indeed, in all methods of "controlled hypotension." The introduction of the methonium compounds, in 1948,[5] provided a simple method of sympathetic blockade at the ganglionic level, merely by injection, and led to the widespread use of the technique of induced hypotension. More recently, Arfonad (d-3,4 [1′,3′-dibenzyl-2′-keto-imidazolido]-1,2 trimethylene thiophanium d-camphor sulfonate), a thiophanium compound, and, to a less extent, Pendiomid (N, N, N′, N′-3-pentamethyl-N, N′-diethyl-3-aza-pentane-1,5-diammonium dibromide), have made possible more readily controllable forms of ganglionic blockade for the purpose of reducing excessive bleeding during operation,[6] again employed in combination with proper positioning of the patient.

Concern naturally arises for the adequacy of circulation to the vital organs during the period of hypotension, in particular to the brain, the heart, the liver, and the kidneys. It has been argued that the level of the systemic arterial pressure is immaterial, as long as there is adequate capillary circulation; this is insured when the pressure at the arteriolar end of the capillary exceeds the sum of the venous pressure and the colloid osmotic pressure of the plasma, a total of about 32 mm. Hg in normal man.[7] Arterial pressures in excess of 32 mm. Hg are expended solely in overcoming peripheral resistance, and when this factor is removed, a systolic pressure somewhat in excess of 32 mm. Hg will provide adequate tissue oxygenation.[8]

This statement requires modification in reference to the cerebral circulation, with the patient in an upright posture, because of the hydrostatic force of the column of blood between the heart and the vertex of the skull. This hydrostatic force may amount to 30 mm. Hg and must be overcome before arterial flow can occur. Hence, for the patient in upright posture, the critical systolic pressure approaches 70 mm. Hg.[8] Cerebral blood flows in three patients studied by Stone following administration of hexamethonium chloride were unchanged in spite of reductions in mean arterial pressure of 40 to 50%.[9] In each instance there was a decrease in cerebrovascular resistance to compensate for the decrease in pressure head. All three patients were young adults. It seems reasonable to assume that different findings may be obtained when these studies are extended to elderly patients with cerebrovascular sclerosis.

5. Barlow, R. B., and Ing, H. R.: Curare-Like Action of Polymethylene bis-Quaternary Ammonium Salts, Nature, London **161**:718 (May 8) 1948. Paton, W. D. M., and Zaimis, E. J.: Curare-Like Action of Polymethylene bis-Quaternary Ammonium Salts, Nature, London **161**: 718-719 (May 8) 1948.

6. Sarnoff, S. J.; Goodale, W. T., and Sarnoff, L. C.: Graded Reduction of Arterial Pressure in Man by Means of a Thiophanium Derivative (Ro 2-2222): Preliminary Observations on Its Effect in Acute Pulmonary Edema, Circulation **6**:63-73 (July) 1952. Nicholson, M. J.; Sarnoff, S. J., and Crehan, J. P.: The Intravenous Use of a Thiophanium Derivative (Arfonad-Ro 2-2222) for the Production of Flexible and Rapidly Reversible Hypotension During Surgery, Anesthesiology **14**:215-225 (May) 1953. Little, D. M., Jr.; Hampton, L. J., and Grosskreutz, D. C.: The Use of 'Arfonad' in a Modification of the Technique of Controlled Hypotension, to be published. Guiot, G., and Damoiseau, F.: Vers la suppression des hemorragies et de l'oedème cérébral par l'emploi des ganglioplégiques eu neuro-chirurgie, Anesthesie et Analgesie **8**:641-648 (Dec.) 1951.

7. Landis, E. M.: Micro-Injection Studies of Capillary Blood Pressure in Human Skin, Heart **15**:209-228 (May 30) 1930.

8. Asquith, E.: Controlled Hypotension in Surgery, Canad. M. J. **67**:421-424 (Nov.) 1952.

9. Stone, H. H.: Personal communication to the authors.

Establishment of a critical systolic level for the patient to undergo operation in the upright position by a similar method of measurement of cerebral blood flows would provide highly desirable and needed information. Investigation of the flicker-fusion intensity in 15 patients following operations performed with "controlled hypotension" suggests that the cerebral circulation is not always sufficient to prevent minor degrees of anoxia, particularly in the head-up position.[10]

Doubt has been expressed concerning the adequacy of the coronary circulation, particularly in older patients subjected to this technique; but no electrocardiographic signs of coronary ischemia were found in either young or old patients with pressures reduced to 60 or 70 mm. Hg.[11]

It has been suggested, as stated above, that a systolic pressure of 32 mm. Hg is the critical level below which capillary circulation cannot be maintained, and there is reason to believe that this figure should be revised upward in postures where hydrostatic pressures are important. In addition, exception must be made for the liver whose blood supply comes from both arterial and venous channels, and where there is danger of anoxic damage, with release of vasodepressor material, if the blood pressure falls below 60 mm. Hg.[12]

Renal filtration apparently is not seriously impaired if the systemic arterial pressure remains above 60 mm. Hg, and, in patients with normal renal vascular channels, kidney damage need not occur in the absence of filtration, provided renal tissue is not made anoxic. Renal circulation is not greatly reduced during controlled hypotension,[13] although anesthesia itself may lower it considerably.[13a]

Because complications of "controlled hypotension" were to be expected and yet, owing to the enthusiasm of proponents of the technique, were not being emphasized, questionnaires were sent to 602 members of the Association of Anaesthetists of Great Britain and Ireland, in an attempt to obtain completely anonymous, but unbiased, reports of the results of the technique, most particularly those fatal and nonfatal complications that had been encountered in its use.[14] Of these questionnaires, 299 (48.06%) were completed and returned, with 178 anesthetists reporting the use of the technique in over 21,000 cases. There were 46 mortalities reported, of which 42 were probably associated with the hypotensive procedure, giving an incidence of 1 death in 500 cases. In many instances there were insufficient data to give true evaluation to the role played by disease and operative trauma in the causation of these deaths. Nonfatal complications occurred 549 times in this series of cases and were related mainly to the cerebral, coronary, and renal circulations.[14]

Similar questionnaires were mailed to the Diplomates of the American Board of Anesthesiology, and the results form the basis of this report. A total of 576 of the

10. Nilsson, E.: The Application of a Method for the Investigation of Cerebral Damage Following Anaesthesia Using Controlled Hypotension: A Preliminary Communication, Brit. J. Anaesth. **25**:24-32 (Jan.) 1953.

11. Enderby, G. E. H., and Davison, M. H. A.: Discussion on the Use of Hypotensive Drugs in Surgery, Proc. Roy. Soc. Med. **44**:829-840 (Oct.) 1951.

12. Bromage, P. R.: Effect of Induced Vascular Hypotension on the Liver: Alterations in Appearance and Consistence, Lancet **2**:10-12 (July 5) 1952.

13. (*a*) Miles, B. E.; de Wardener, H. E.; Churchill-Davidson, H. C., and Wylie, W. D.: The Effect on the Renal Circulation of Pentamethonium Bromide During Anaesthesia, Clin. Sc. **11**:73-79 (Feb.) 1952. (*b*) McQueen, E. G.: Hexamethonium Bromide and Kidney Function, M. J. Australia **1**:769-771 (June 7) 1952.

14. Hampton, L. J., and Little, D. M., Jr.: The Results of a Questionnaire Concerning the Use of Controlled Hypotension in Anesthesia, Lancet, to be published.

851 questionnaires which were sent have been completed and returned. This constitutes a response of 67.7%. Improper wording of certain questions has resulted in an accumulation of much data that has no statistical value, although it does give intriguing information regarding the clinical practice of "controlled hypotension." There were 144 anesthesiologists who reported the use of such procedures in 6,805 patients.

The techniques that were employed are listed in Table 1.

In regard to the levels of systolic pressure considered necessary for the control of bleeding, 98 anesthesiologists, reporting 3,083 cases, sought a value under 80 mm. Hg, whereas 30 anesthesiologists reporting 2,667 cases sought a level of 80 mm. Hg or above. This represents a more conservative attitude than that displayed by the anesthetists of Great Britain and Ireland, since some 17,000 of the 21,000 cases which they reported were listed in the "under 80 mm. Hg" group.

The use of vasopressors to return the blood pressure toward normal at the conclusion of operation is indicated in Table 2. Owing to poor wording of the question-

TABLE 1.—*Techniques Employed to Obtain "Controlled Hypotension"*

	Anesthesiologists, No.	Cases, No.
Arteriotomy	12	208
Spinal	46	2,655
Methonium compounds	115	3,375
Thiophanium compounds	16	359
Pendiomid	6	63
Others	14	145
Total		6,805

TABLE 2.—*Number of Anesthesiologists Employing Vasopressor Agents to Restore the Blood Pressure Toward Normal in "Controlled Hypotension"*

Yes	63
No	28
Sometimes	45

naire, it is not known how many anesthesiologists used vasopressors routinely and whether they used such drugs before the surgical wound was closed or later. The complication of reactionary hemorrhage occurred 10 times in the group in which vasopressors were used "when necessary," as contrasted with 49 times in the group in which vasopressors were used as a routine.

"Controlled hypotension" was employed chiefly in neurosurgical procedures, especially craniotomies, and less frequently in tumor surgery involving radical dissections of the head and neck, the chest wall, the lower abdomen, and the pelvis. Only one anesthesiologist mentioned plastic surgery as an indication for the technique (Table 3). Five, including us, used it in surgery involving the great vessels in the mediastinum to decrease tension and thereby facilitate the placing of clamps and suture lines.

The suggested contraindications to these techniques included disease of various branches of the vascular tree, hepatic and renal disease, blood volume disturbances, and uncomplicated elective surgery (Table 4).

The complications reported are listed in Table 5. These were then divided into two groups: those reported by anesthesiologists who strove to obtain systolic blood pressures of less than 80 mm. Hg, and those reported by anesthesiologists who sought

TABLE 3.—*Type of Procedures in Which "Controlled Hypotension" was Employed*

	Anesthesiologists
Craniotomy	87
Neurosurgical, unspecified	30
Head and neck	25
Chest-wall (mastectomy)	19
Intrathoracic	12
Cardiovascular	5
Abdominal	20
Pelvic (radical)	31
Orthopedic	8
Plastic	1

TABLE 4.—*Contraindications to the Use of "Controlled Hypotension"*

		Anesthesiologists
Cardiovascular disease		175
Coronary disease	57	
Cardiac disease	45	
Arteriosclerosis	30	
Cardiovascular disease	19	
Severe hypertension	24	
Cerebrovascular disease		12
Hepatic disease		14
Renal disease		40
Age over 50 to 60 yr		25
Blood-volume disturbances		60
Anemia	18	
Hemorrhage	7	
Shock	12	
Low blood volume	19	
Polycythemia	4	
Uncomplicated elective surgery		17
Others		59

TABLE 5.—*Complications Encountered in 6,805 Cases of "Controlled Hypotension"*

	Cases, No.
Anuria	8
Oliguria	24
Cerebral thrombosis	9
Retinal thrombosis	1
Coronary thrombosis	3
Cardiac arrest	15
Cardiovascular collapse	25
Reactionary hemorrhage	79 (25 mild)
Persistent hypotension	70
Delayed awakening	3
Others	17

a pressure no lower than 80 mm. Hg. These figures were subjected to statistical analysis employing the chi-square test, and the results are tabulated in Table 6. It may be concluded from these statistics that one method of decreasing the number of complications associated with "controlled hypotension" lies in maintaining the systolic blood pressure at a level of 80 mm. Hg or above.

Fifty deaths were reported. Of 25 deaths which followed craniotomies, 11 involved ligation of aneurysms. Inspection of Table 7 shows how difficult it is to demonstrate cause-and-effect relationship between "controlled hypotension" per se and the stated cause of death.

SUMMARY

The results of questionnaires submitted to members of the Association of Anesthetists of Great Britain and Ireland and to Diplomates of the American Board of Anesthesiology reveal that "controlled hypotension" has been employed in 28,000 cases and that its use is accompanied by certain complications which are inherent in the technique itself.

Results of the American questionnaire contain data of statistical significance which indicate that in inducing hypotension 80 mm. Hg is the critical level of systolic blood pressure above which complications are infrequent.

This is not an indictment of "controlled hypotension." When wisely employed in the carefully selected case for an urgent and rational indication, the technique may prove markedly beneficial.

Drs. Georgina Y. Goodwin and Margrethe J. Kingsley, residents in anesthesiology, gave invaluable aid in the tabulation of the figures.

TABLE 6.—*Comparison of Blood Pressure Levels with Occurrence of Complications in "Controlled Hypotension"*

	Below 80 Mm. Hg (3,071 Cases)	80 Mm. Hg or More (2,664 Cases)	P	
Anuria	7	0	0.05>P>0.02	? Significant
Oliguria	17	5	0.05>P>0.02	? Significant
Cerebral thrombosis	9	0	0.02>P>0.01	Significant
Retinal thrombosis	1	0		
Delayed awakening	2	0		
Coronary thrombosis	2	1		
Cardiac arrest	14	1	0.01>P>0.001	Highly significant
Cardiovascular collapse	25	0	0.01>P>0.001	Highly significant
Reactionary hemorrhage	71	7	0.01>P>0.001	Highly significant
Persistent hypotension	53	7	0.01>P>0.001	Highly significant

TABLE 7.—*Deaths Occurring in 6,805 Cases of "Controlled Hypotension"*

Cause of Death	Cases, No.
Anuria	1
Cerebral thrombosis	4
Cerebral edema	3
Coronary thrombosis	1
Cardiac arrest	2
Cardiovascular collapse	4
Postoperative hemorrhage	6
Persistent hypotension	1
Arterial air embolism	1
Aortic-femoral thrombosis	1
Pulmonary infarcts, edema	2
Following high spinal anesthesia	3
Neurosurgical operative trauma	14
Peritonitis	2
Carcinoma with metastases	3
Preexisting cardiovascular disease	1
Atelectasis	1

ABSTRACT OF DISCUSSION

DR. STEVENS J. MARTIN, Hartford, Conn.: The authors have presented briefly the historic development of anesthetic techniques with "induced hypotension," emphasizing the complications associated with their use. Like other new anesthetic techniques or agents, their introduction is accompanied by an initial wave of enthusiasm, followed by a wave of condemnation, and finally, after more intensive laboratory and clinical evaluation, there is a wave of acceptance. It appears that the authors have attempted to wade through the wave of condemnation and to point the way to accepting anesthetic techniques with "controlled hypotension," but with certain reservations.

The data obtained from a questionnaire sent to many competent anesthesiologists in the world has not been submitted to a thorough statistical study. Therefore no conclusion per se may be made. However, the data are important in some of their specific details.

First, the paper emphasizes some precautions, limitations, contraindications, and complications incident to anesthesia with controlled hypotensive technique. This constitutes a major step in evaluating this new procedure for clinical use. The incidence of morbidity and mortality, even in competent hands, indicates that hypotensive techniques must not be used indiscriminately, but in very carefully selected cases. Furthermore, it should be emphasized that these techniques should not be employed as a convenience to the surgeon, but rather as an aid to the patient.

It is regretted that the authors made no attempt to make specific correlations between complications seen in controlled hypotensive anesthesia due to (1) the reduction of a patient's blood volume (arteriotomy or phlebotomy) or those due to (2) the decrease in peripheral resistance (high-level spinal anesthesia, postural pooling, deep anesthesia, autonomic ganglia blockade at the fifth or sixth cervical level, or to combination of these methods). In arteriotomy there is a compensatory increase in arteriolar tone, with a decrease in tissue blood flow and oxygenation. With decrease in peripheral resistance there is presumably adequate tissue blood flow and oxygenation.

Secondly, has any anesthesiologist completing his questionnaire studied the following data on his patients: (*a*) electrocardiograms, (*b*) circulation time, (*c*) venous pressure, (*d*) auriculoventricular oxygen differences of the brain, (*e*) oximeter readings, and (*f*) electroencephalograms. Too few of these very significant correlations have been published.

Lastly, the most important feature of this paper is its provocative current interest. Aside from studies already reported by surgeons, controlled hypotensive anesthesia has been discussed in written and oral examinations given to prospective diplomates of the American Board of Surgery. This paper should prove to be most interesting to our surgical confreres. In listening to the paper presented today, many in the audience may feel prompted to ask the following questions: 1. Why did 17 anesthesiologists participating in the questionnaire indicate "uncomplicated elective surgery" as a contraindication to controlled hypotensive anesthesia? 2. Is the incidence of complications and of deaths compatible with that seen with established well-known anesthetic techniques? 3. There are only meager data on cerebral blood flows during controlled hypotensive anesthesia. Has any one in the questionnaire group made such studies? 4. Lastly, on the basis of the questionnaire and personal experiences, would the author kindly discuss briefly the indications of controlled hypotension during anesthesia?

DR. SCOTT M. SMITH, Salt Lake City: I think that this study and survey certainly presented very timely information. I believe that Dr. Martin has asked many of the questions that have come to our minds. I think we are grateful to Dr. Hampton for the attempt to collect and evaluate material that is available but is very difficult to get (and even once obtained, the analysis is indeed difficult).

One of the things I should like to see done is a comparison of the mortality and morbidity of the patients undergoing comparable procedures without the use of hypotensive agents. I believe that some neurosurgeons for certain neurosurgical procedures probably would be happy to have no more than 50% mortality. I should like to hear more about the indications. I am sure we are going to use it in some instances for the convenience of the surgeon, if it does not increase the hazard to the patient, and I think we would be justified in doing so because part of our survival depends upon doing things for the convenience of the surgeon, as long as it does not increase the hazard to the patient.

Dr. C. Paul Boyan, New York: I have been using hypotensive anesthesia for the past two years at Memorial Hospital. We use this type of anesthesia for radical surgical procedures in the pelvis and in the abdomen. I was not prepared to talk today, consequently I can not give figures, but I can tell you roughly. I can also answer one of the questions Dr. Martin brought up. When radical surgery is attempted, the bleeding is sometimes very profuse; we have lost patients on the table because of the bleeding. We replace the blood volume, but the patients do not do too well, and we give more blood to them and there is more bleeding. We have tried all kinds of drugs, and we have tried fresh plasma and 30 kinds of blood. You see, that is one of the dangers of radical surgery.

This afternoon I am going to report on a series of procedures done with use of this type of anesthesia. In these cases we have not had any deaths on the table due to bleeding. However, if we compare 111 cases of major surgery in which normal anesthesia was used, there will surely be a few deaths. I agree with the neurosurgeons that they have good indications for their use of hypotensive anesthesia, although they are somewhat handicapped in having the patient in that position. When this type of anesthesia was first used in England, the physicians were extremely enthusiastic. They were giving hypotensive anesthesia for tonsillectomy, for tooth extraction, for practically everything, but after a while they discovered complications, and now they are somewhat more reserved, as are we.

Dr. L. Jennings Hampton, New Haven, Conn.: In regard to the first question raised by Dr. Martin, the comparison of complications arising from arteriotomy with those of other methods, this would be interesting to do because the techniques of physiology are somewhat different. Unfortunately, on the questionnaires which we used, the persons were asked to state the types of techniques used, and they may have included some cases without and some with blockade. We cannot break it down as to which occurred under which technique, consequently we cannot assemble such data.

I do not know why some persons put down "uncomplicated elective surgery," except that perhaps they had in mind such items as the death that was reported by one of our visitors from Great Britain last fall, in a case in which he set out to accomplish, and did accomplish, a blockade; the procedure was with the face and feet with cardiac rest, and the patient did not respond to treatment. Unless we feel we have rather definite indications, we are not in a position to advocate this method without qualification.

The question regarding the relative incidence of morbidity and mortality in comparable procedures with hypotension or without hypotension is a valid point to emphasize because, particularly in the questionnaire circulated here, it was apparent that the technique was being used for a great number of procedures which have always carried a very high incidence of complications and mortality, for example, ligation superannulus.

As to the studies of cerebral blood flow, the only ones I know of are those being done by Dr. Stone, in Philadelphia, but undoubtedly there are others.

One more word about the cerebral blood flow. If we are going to assume one of the indications for this type of technique in surgery, much of which is done best with the patient in a head-up posture, if not erect posture, it is very important that we know at what level and in what position cerebral flow is seriously impaired.

The indications that we feel are uppermost are intracranial surgery, secondary to the one that Dr. Boyan mentioned, the valuable tumor surgery involving great areas of resection, and surgery in which cleavage may not be too well established and which also carries with it the danger of hemorrhage.

Those performing cardiovascular surgery, particularly ligation and coarctation and excision of thoracic aneurysm, will find a modified type of hypotension is extremely valuable. It serves to decrease the tension in the wall and makes it much more feasible to place clamps, and I think, instead of adding to the complications, will probably decrease the number of complications associated with that type of surgery. The British have used the plastic surgery, some of which involves a great deal of bleeding, and we were rather surprised when only one person who answered the questionnaire marked "plastic surgery" as an indication. I am sure I have others, but time is short.

SIMULTANEOUS QUADRILATERAL GANGRENE

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SIMULTANEOUS quadrilateral gangrene is a rare finding. We are prompted to report this syndrome, since in our total experience we have never previously observed it. Review of the literature reveals a paucity of case reports. These were observed in frost bite, thromboangiitis obliterans, carcinoma, cryoglobulinemia, and endocarditis. A case of hypothermia with quadrilateral gangrene simultaneously was reported by Laufman.[1] Previously Theis and co-workers[2] reported another case of quadrilateral frost-bite gangrene. Luckey and his associates reported the unusual occurrence of simultaneous quadrilateral gangrene in cryoglobulinemia.[3] A case of quadrilateral gangrene associated with carcinoma of the pancreas and acute nonbacterial endocarditis was reported.[4] There are no references in the literature to simultaneous quadrilateral gangrene in thromboangiitis obliterans, but quadrilateral gangrene has been reported.[5]

Symmetrical gangrene of the lower extremities has been reported frequently. In children it has been noted by Lewis[6] and Richter.[7] Broders and Snell have

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1. Laufman, H.: Profound Accidental Hypothermia, J. A. M. A. **147**:1201 (Nov. 24) 1951.

2. Theis, F. V.; O'Connor, W. R., and Wahl, F. J.: Anticoagulants in Acute Frostbite, J. A. M. A. **146**:992 (July 14) 1951.

3. Luckey, E. H.; Russ, E. M., and Barr, D. P.: Cryoglobulinemia: Influence of a Cryoglobulin on the Suspension Stability and Sedimentation Rate of Erythrocytes, J. Lab. & Clin. Med. **37**:253 (Feb.) 1951.

4. Clinicorathologic Conference, Brooklyn Hosp. J. **4**:53 (July) 1946.

5. Collens, W. S., and Wilensky, N. D.: Peripheral Vascular Diseases: Diagnosis and Treatment, Ed. 2, Springfield, Ill., Charles C. Thomas, Publisher, 1953, p. 120. Allen, E. V.; Barker, N. W., and Hines, E. A., Jr.: Peripheral Vascular Diseases, Philadelphia, W. B. Saunders Company, 1946, p. 447. Thromboangiitis Obliterans, Foreign Letters, J. A. M. A. **150**:1716 (Dec. 27) 1952.

6. Lewis, T., and Pickering, G. W.: Observations upon Maladies in Which Blood Supply to Digits Ceases Intermittently or Permanently, and upon Bilateral Gangrene of Digits: Observations Relevant to So-Called "Raynaud's Disease," Clin. Sc. **1**:327 (Dec.) 1934; in Cecil, R. L., Editor: Textbook of Medicine, Ed. 4, Philadelphia, W. B. Saunders Company, 1937, p. 1128.

7. Richter, I. H., and Tainsky, I. A.: Gangrene of the Lower Extremities in Infants: Report of 2 Cases, Am. Heart J. **36**:443 (Sept.) 1948.

reviewed the literature of fulminating meningococcemia with gangrene.[8] Cold hemagglutinins have produced symmetrical gangrene of the hands.[9] Yater and Cahill have reported bilateral gangrene of the feet due to ergotamine tartrate.[10] Multiple arterial thrombosis has been seen simultaneously to involve both the lower extremities and the right arm.[11] In malaria, symmetrical gangrene of the lower extremities has been reported.[12] Likewise, symmetrical gangrene has been noted in Raynaud's disease.[13] We have reported bilateral simultaneous gangrene of the lower extremities with recurrent thrombophlebitis in obscure carcinoma of the lung.[14] We (Fisher and Wilensky) are reporting bilateral simultaneous gangrene of the lower extremities following a cut-down for an intravenous infusion. In acute infections such as scarlet fever[15] and diphtheria,[16] gangrene of the lower extremities has been reported. Dr. Lord has observed two cases of simultaneous quadrilateral gangrene at Bellevue Hospital Center, New York.

The above observations, derived from the literature, are all infrequent. On occasion, bilateral symmetrical gangrene is seen in cases of saddle thrombus of the aorta. Bilateral symmetrical lesions can also be seen in cases of peripheral arteriosclerosis obliterans of the lower extremities. It has also been seen in patients with the Leriche syndrome.

REPORT OF CASES

CASE 1.—C. M., aged 68, was admitted to the Kings County Hospital Jan. 26, 1953, with complaints of pain and blueness of the fingers of five weeks' duration. About five weeks before admission the patient noted numbness and tingling of all the digits of his hands. Soon thereafter the fingers became reddened and warm, then blue and cold. No pain was noted at that time. The patient stated that for many years previously his fingers would turn blue and cold in cold weather. One week after the onset of this illness the patient's ankles became swollen, and dyspnea developed upon exertion. The blueness of the fingers persisted, and moderate pain developed until the time of admission.

The patient had been in previous good health, except for an episode of tuberculosis 30 years previously. He had had a chronic nonproductive cough for many years, which he ascribed to excessive smoking. He had smoked two packs of cigarettes daily for over 20 years. He stopped smoking two months prior to admission. He had had a poor appetite and loss of weight for the

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9. Stats, D., and Bullowa, J. G. M.: Cold Hemagglutination with Symmetric Gangrene of the Tips of the Extremities: Report of a Case, Arch. Int. Med. **72**:506 (Oct.) 1943.

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13. Raynaud, A. G. M.: De l'asphyxie locale de la gangrène symétrique des extrémitiés, Paris, Rignoux, 1862, p. 115.

14. Fisher, M. M.; Hochberg, L. A., and Wilensky, N. D.: Recurrent Thrombophlebitis in Obscure Malignant Tumor of the Lung: Report of 4 Cases, J. A. M. A. **147**:1213 (Nov. 24) 1951.

15. Learmonth, B. E.: Gangrene of the Lower Extremities Complicating Scarlet Fever, Canad. M. A. J. **15**:69 (Jan.) 1925.

16. Bailly, L. A.: Occlusion of Arteries of Limbs in Diphtheria, Interna. Clin. **2**:157, 1920.

past 10 years, which became severer in the past 2 years. There had been no change in bowel habits. The patient denied any history of excessive alcoholic consumption. He had not been taking any ergot drugs or any other medications. He had not been exposed to excessive cold or chemical fumes. He had not eaten any spoiled rye bread. The patient had been employed as a Wall Street runner until the onset of his illness.

Examination revealed a pale slender white man in no apparent distress. He was alert and cooperative. Blood pressure was 168/80 mm. Hg; pulse rate 100 per minute; respiration 18 per minute, and temperature 100.2 F. The chest was symmetrical and clear to percussion and auscultation. The heart was not enlarged to percussion and had a regular sinus rhythm. No murmurs were heard. The abdomen revealed no masses or tenderness; liver, kidney, and spleen were not palpable. All the fingers of the hands were cold and cyanotic up to midway between the first and second interphalangeal joints. No clubbing of the fingers was present. There was a bilateral 2 + pitting edema of the ankles. The toes and soles of the feet were cold and cyanotic. A delayed capillary filling time was noted on clenching of the fists. Peripheral pulsations were present in all extremities. Oscillometric readings were normal throughout. Deep tendon reflexes were equal bilaterally. No pathologic reflexes were elicited. Pain, touch, and temperature sensation were absent over all cyanotic areas. The cranial nerves were intact. Motor power was present in all extremities. Fasciculations were noted in the muscles of the upper extremities.

Laboratory Data.—The hemoglobin on admission was 13 gm.; the white blood cell count was 9,200; the urine was negative. A repeat blood study one week after admission revealed a hemoglobin of 9.7 gm.; red blood cell count 3,050,000; white blood cell count 13,700, and the differential count 81% neutrophiles, 3% bands, 7% lymphocytes, 9% monocytes. There were 567,000 platelets. Blood chemistry studies revealed the following values: cephalin flocculation 2+, total serum cholesteral 116 mg., total protein 7.0 gm., albumin-globulin ratio 3.7 : 3.3, icteric index 3 units, alkaline phosphatase 0.4 mg., serum amylase 240 units, serum phosphates 5.5 mg., serum calcium 8.4 mg. A test for cold agglutination was positive in a 1 : 4 dilution on one occasion and negative on three subsequent occasions. Tests for serum cryoglobulins were negative on three occasions. The Wassermann reaction of the blood was negative. X-ray studies of the hands showed no bone or joint pathology. An upper gastrointestinal series was negative. Chest x-ray revealed a moderate-sized pleural effusion on the left, with obscuration of the lower half of the left lung field and poor aeration of the remaining visualized lung field. Marked interstitial fibrotic changes were noted in the contralateral lung field.

Soon after the demonstration of fluid on x-ray, a thoracentesis was performed on the left lung, and 300 cc. of straw-colored fluid was aspirated. This fluid was negative for malignant cells. Bronchoscopy revealed normal orifices in the bronchus of the upper lobe of the left lung. Some inflammation and edema of the lower lobe orifices were noted. A culture of the bronchial washings was reported as positive for acid-fast bacilli.

The soles and toes of the feet became increasingly cyanotic. Two bullae formed on the dorsum of the left foot, together with a cellulitis of the two feet. The patient's temperature at this time was 100 F. He was placed on penicillin, heparin sodium (Depo-Solution Heparin Sodium), and Priscoline (2-benzyl-4,5-imidazoline hydrochloride). The hands and feet were wrapped in cotton. Paravertebral blocks of the bilateral stellate ganglia were attempted, with no change in the condition of the extremities. A histamine wheal test done at this time was negative. By the end of the second week the patient's digits had become completely mummified. There was no pain present. The gangrenous areas on the hands had become well demarcated at a point midway between the middle and distal interphalangeal joints (Figs. 1 and 2). The toes were completely gangrenous, as were large areas of the soles of the feet (Fig. 3). The patient's general condition deteriorated. He became markedly emaciated and lethargic and died on the 23rd hospital day.

Autopsy Report.—Following are the relevant gross finding at autopsy, performed by Dr. Harry Iticovici. The body of an elderly, poorly nourished, slightly built man showed evidence of a dry gangrenous process symmetrically and equally distributed involving the extremities of all limbs. The line of demarcation was irregular but sharp; the digits were shriveled, blue-black, and dry. The hands were involved proximally as far as the first interphalangeal joint of each

finger, except for the thumb, where the discloration extended only over the distal phalanx. The feet were similarly affected over an area which encompassed the entire surfaces, including the heels, the instep on the right foot, all the toes, and the dorsum of the two feet as far proximally as the metacarpophalangeal joints on the right foot and the distal margin of the astragalus on the left foot. Proximal to the margins of gangrene, small areas of erythema (4 by 3 cm.) with excoriation of the skin were noted. A decubitus ulcer covered with a gray-black scab was present in the sacral region. A 5 by 10 cm. ecchymotic area around the left elbow and similar

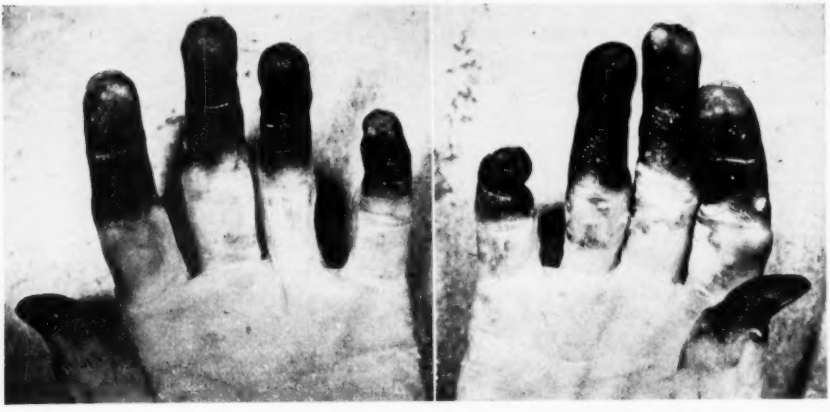

Fig. 1 (Case 1).—Palmar aspect of the patient's hands.

Fig. 2 (Case 1).—Dorsum of the patient's hands.

discolorations of the hands were present. Elsewhere the skin was white and flabby and showed poor turgor. The muscles of the palms were red-brown and flabby.

Segments of the following vessels of the upper extremities were examined and portions removed for sections; radial, ulnar, superficial and deep palmar arches, and digital. The same was done bilaterally for the dorsalis pedis, posterior tibial, plantar arteries, and digital vessels of the feet. All these vessels, both arteries and veins, seemed patent, soft, and collapsible; no gross anomalies were noted. On milking the iliac veins, no thrombi were expressed. The iliac arteries were devoid of arteriosclerotic plaques and showed no occlusion. The aorta exhibited occasional elevated nonulcerated yellow-white hardened plaques evenly scattered throughout its

length and not occluding any orifice of its branches. This intimal process involved approximately 10% of the aortic internal surface area and less severely affected its major branches. The cerebral arteries showed minimal yellowish thickening.

The heart weighed 200 gm. and was normal in shape, in all dimensions, and in appearance of the myocardium. The aortic valve cusps were somewhat brittle, hardened, and yellow. The mitral valve showed a few small hard yellow nodules on the leaflet. The coronary arteries, sectioned serially, were soft and collapsible and revealed no occlusive lesions.

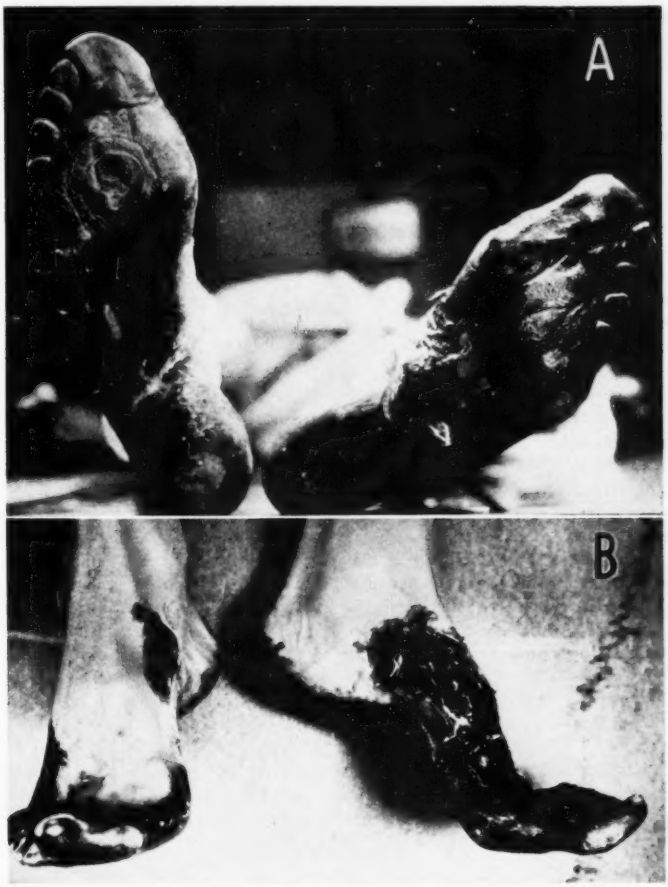

Fig. 3 (Case 1).—*A*, plantar aspect of the patient's feet. *B*, dorsum of the patient's feet.

Each lung weighed 450 gm. The upper lobe of the right lung was firmly adherent to the parietal pleura and was covered with many fibrous adhesions, which also bridged the interlobar fissures. Its substance contained white streaks of tissue posteriorly; whereas the apex itself was consolidated and presented a creamy doughy cut surface. Some frothy fluid could be expressed from a reddened lower lobe. The left lung was collapsed to approximately 75% of its normal volume, and the left pleural cavity contained approximately 500 cc. of straw-covered fluid. Some fibrous adhesions were present over the apex and along the mediastinal surface. The two lobes

were noncrepitant and presented a solid red cross section studded with purulent gray material. The tracheobronchial lymph nodes were discrete and black, measuring up to 1 cm. on cross section.

The remainder of the organs were within normal limits for the man's age and presented nothing of note, except some slight pitted scarring of the renal surfaces and a mild nodular enlargement of the prostate.

Microscopic Findings.—The lungs were the seat of a widespread and extensive inflammatory process, both old and recent, inactive and active tuberculosis. Many areas of plumonic parenchyma were markedly altered by extensive hyaline fibrosis mostly in coarse anastomosing bands with conspicuous deposition of anthracotic pigment. Many poorly cellular fibrous nodules with amorphous granular central areas were intimately associated with the more diffuse variety of fibrosis. Similar old tuberculoid granulomata occupied some of the hilar lymph nodes. In some areas, particularly in the vicinity of some of the more active granulomatous lesions, the aerated portions of lung displayed a more diffuse but less severe and more cellular variety of fibrosis of the alveolar septa, accompanied by a scattered lymphocytic infiltration. There was plugging of some of the air sacs with loose fibrous tissue plugs attached to the surrounding alveolar walls. Elsewhere the air spaces were moderately filled with large and small mononuclear cells, the larger ones containing appreciable quantities of sudanophilic droplets. Other areas of lung, particularly corresponding to the upper lobe of the right lung, revealed a much more exudative process, the predominant theme of which was caseation necrosis. Where this was more self-contained, focal, and obviously granulomatous, epithelioid cells, lymphocytes, and Langhans' giant cells were evident in the usual arrangement; elsewhere the process overran all normal structure, sometimes obliterating it in a mass of caseous necrotic material. In these areas the Ziehl-Neelsen stain showed large numbers of acid-fast beaded rods identifiable as Salmonella typhosa. A second process, a suppurative one, was noted to impinge upon and modify the tuberculous inflammation. In some areas this was the sole lesion involving pulmonary tissue, and its bronchopneumonic distribution was evident; in others, where it was mingled with areas of ceaseation, it tended to produce irregular abscess-like cavities partially filled with polymorphonuclear leucocytic nuclear elements in an amorphous mass. Few and small were regions of relatively normal pulmonary parenchyma.

Sections of arteries of all sizes revealed a minimal degree of uneven intimal widening, with fairly cellular fibrous tissue containing little or no sudanophilic material. This widening, for the most part, did not encroach appreciably on the width of the lumina and was not of the typically atheromatous variety; it was more cellular in the muscular arteries and of a more hyaline quality in the aortic intima. Some reduplication of the internal elastic membrane was noted frequently, with minimal or absent calcification of the Mönckeberg type. Only two instances of occlusive lesions were found. One of these involved an artery approximately 6 mm. in outside diameter. The arterial wall was markedly and unevenly fibrosed and calcified mainly in the intimal and inner muscular portions, this fibrosis blending imperceptibly with a mass of loose fibrous tissue which filled the lumen. The second involved a small artery approximately 1 mm. in outside diameter (Fig. 4). This artery showed changes similar to those described for the majority of the arteries; its lumen, moreover, was filled with a moderately cellular fibrous tissue which was recanalized and contained a few foci of brown hemosiderin-like pigment. The vasa vasorum were somewhat prominent in a slightly fibrosed adventitia. No granulomata of any sort were noted. The veins and nerves were within normal limits; no thromboangiitis obliterans was noted in the veins and arteries.

Sections of skin at the margin of the gangrenous process reflected a coagulative process superficially replacing the epidermis with a necrotic sloughing layered mass of tissue showing polymorphonuclear leucocytic infiltration. In the vicinity of these areas an occasional arteriole was noted to be plugged with a finely granular, partially hyaline mass attached to the wall and covered with endothelium. Striated muscle in foci revealed evidence of loss of striations, mild swelling, or atrophy, accompanied by mild mononuclear cell infiltration; in some areas it showed replacement by amorphous masses of eosinophilic tissue which stained negatively for amyloid material and tended to retain the outline of the muscle it replaced.

A moderate increase in the number of plasma cells in the bone marrow and in the spleen was noted.

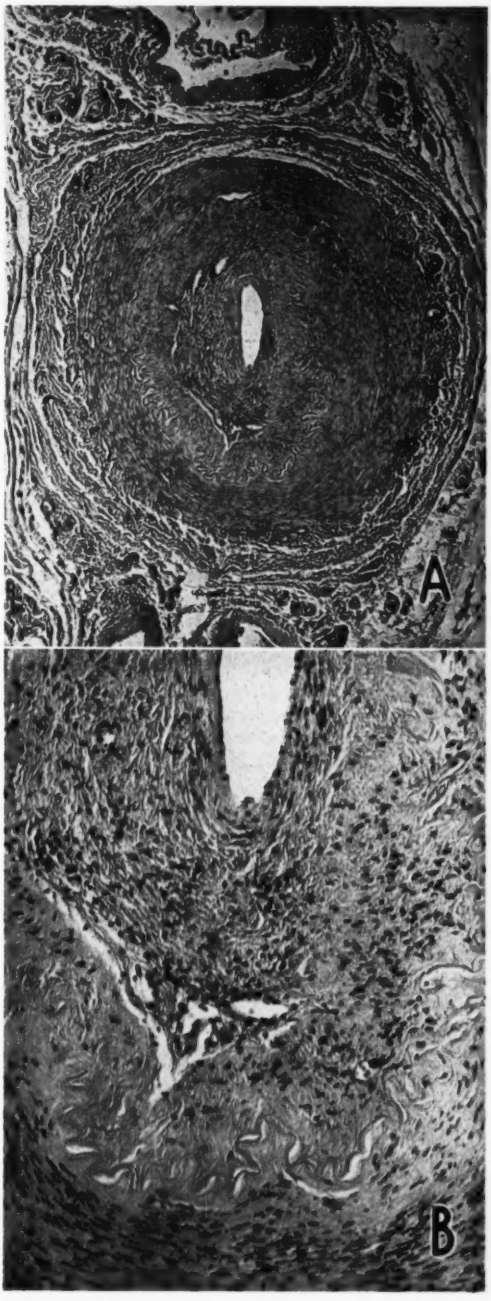

Fig. 4.—Microscopic section through a small artery. Lumen is filled with a moderately cellular fibrous tissue which was recanalized. *A*, × 45. *B*, × 150.

Anatomical Diagnoses.—The anatomical diagnoses were fibrocaseous pulmonary tuberculosis, focal organized pneumonitis, caseous pneumonia (extensive) with superadded suppurative elements, pleural effusion (left), splenitis, gangrene of all extremities, arteriosclerosis (mild) with old thrombosis, chronic pericarditis (mild), nodular hypertrophy, and hyperplasia of prostate.

COMMENT

The tuberculous pneumonic process in this patient indubitably antedated the vascular phenomena and constituted the immediate cause of death, which was further aggravated by the acute bronchopneumonic suppurative disease. Though anatomical evidence is present of arteriosclerosis with occlusion, the mildness of the occlusion and the minimal degree of the arteriosclerosis militate against incriminating anatomically arteriosclerotic occlusive disease for the quadrilateral gangrene, though it may have afforded a suitable background. The age of the patient, the lack of intermittent claudication, and the morphological changes present make a diagnosis of thromboangiitis obliterans essentially untenable. A superadded vasospastic phenomenon is strongly suggested by the unusually extensive and symmetrical distribution of the gangrene. This may have acted synergistically with the pulmonary disease to bring about the final turn of events.

CASE 2.—J. D., aged 68, was admitted to the Kings County Hospital Feb. 23, 1953, complaining of ankle swelling of one month's duration. The patient had been in previous good health, except for arthritis and low blood pressure. One month prior to admission the patient noted intermittent swelling of the ankles. At the same time he also noted that the first and second toes of his left foot had become black.

Examination revealed a well-developed, well-nourished white man in no apparent distress. He was mentally confused and uncooperative. Blood pressure was 110/80 mm. Hg; pulse rate 78 per minute; respiration 12 per minute, and temperature 99 F. No unusual physical findings were noted, except in the lower extremities. There was bilateral ankle edema. Ulcers were present on the heels; gangrene of the first and second left toes was present. All peripheral pulses were easily palpable. Oscillometric readings were normal. No clubbing of the fingers was noted.

Laboratory Data.—The hemoglobin on admission was 12.5 gm.; white blood cell count was 9,000; urine was normal; sedimentation rate was 24 mm. in 20 minutes (Sahli). The following values were given: blood urea nitrogen 11.0 mg.; fasting blood sugar 80 mg.; total serum cholesterol 200 mg.; total serum protein 7.3 gm., with an albumin-globulin ratio of 4.5 : 1.8. The Wassermann test was negative. X-rays of the hands and feet showed demineralization of the bones. A gastrointestinal series revealed a marked deformity of the duodenal cap consistent with a chronic duodenal ulcer. A gall bladder series showed nonvisualization. The chest and skull studies were negative.

The patient was started on a course of tetraethylammonium (Etamon) chloride therapy; despite therapy, however, by the fourth hospital day the gangrene had progressed to involve all four extremities. All of the toes, as well as the fourth and fifth digits of the left hand and the second digit of the right hand, were gangrenous. The patient began having episodes of rectal bleeding and acute psychotic outbreaks. The gangrene progressed, and on the 29th hospital day he went into shock and died. No autopsy was obtained.

COMMENT

Simultaneous quadrilateral gangrene is being reported as a rare observation. We have seen two cases on the wards of the Kings County Hospital in Brooklyn; on one patient an autopsy was performed. From our postmortem protocol it is noted that most of the arterial tree was dissected without any explanation for the clinical findings. All possible etiological factors were considered and were eliminated through the history, laboratory findings, or autopsy findings. Minimal amount of atherosclerosis was seen on the microscopic sections. The cold agglutination tests

were performed and were negative. Cryoglobulinemia was not found. The thrombogenic diseases were ruled out by a normal hemogram. No evidence of malignancy was found. Although the patient suffered from pulmonary tuberculosis, no evidence of tuberculous arteritis was noted on any sections. A careful history obtained from the patient and his family failed to elicit any evidence of exposure to cold, chemical fumes, ergot, or trauma. The serology was negative. The patient was afebrile. Except for the tuberculosis, no other evidence of systemic or focal infection was noted. No evidence of a focus for embolization was found in the heart, large vessels, or pelvic vessels. There was no evidence of essential polyangiitis. There was a history of the Raynaud phenomenon.

The second patient also had quadrilateral gangrene while under our observation. Again no etiological diagnosis was made. Unfortunately we were unable to obtain an autopsy.

SUMMARY

Two cases of simultaneous quadrilateral gangrene are reported. The etiological factors are obscure. The possible causes are discussed and do not help to make a diagnosis.

Cooperation in the preparation of this article was given by Dr. Arthur Fankhauser, Dr. Victor Ginsburg, the Department of Pathology, and Dr. Henry Siegel.

PRIMARY TUMORS OF THE RIB

Review of the Literature and Presentation of Eleven Cases Not Reported Previously

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PRIMARY tumors of the rib, though uncommon as compared with similar tumors arising in other bony structures of the body, are not so rare as has been supposed. Such new growths are recognized more frequently today than in the past because of the improved diagnostic methods and the more complete knowledge on the part of the clinician, radiologist, surgeon, and pathologist of the nature of these lesions. This greater familiarity with these tumors has led to the erroneous impression that the incidence of rib tumors is rising. That this is not the case becomes obvious from a study of the cases reported in the world literature for the last half-century. Such studies also show that all types of primary neoplasms involving bone and cartilage, in other parts of the skeleton, may also be encountered in the ribs.

Though more readily detected in our day than in former years, primary tumors of the rib are still uncommon in any one physician's experience, a fact that sometimes leads to serious errors in diagnosis, prognosis, and treatment. It is with this in view that the present paper seeks to serve a threefold purpose: (1) to assemble and offer in "digest" form relevant data gained from a detailed, though necessarily incomplete, study of the literature on primary rib tumors through 1951; (2) to comment briefly on such basic matters as etiology, incidence, diagnosis, and treatment; and (3) to contribute 11 cases not previously reported.

GENERAL SURVEY OF THE LITERATURE

For many years reports of instances of primary rib tumors have been appearing in the medical journals, and attempts have been made from time to time to collect and classify the reported cases.

In 1897 Paget collected and reported 33 cases of primary tumors of the ribs and sternum. Hedblom, in 1921, collected 213 cases from the literature and 12 years later increased this number to 291. To these he added 22 personal cases.[1] These early efforts were not too helpful in completing the clinical picture of tumors of the bony chest wall, as lack of histologic data about the tumors (essential to their proper classification) and insufficient follow-up made it impossible to evaluate the nature of these tumors and the outcome of treatment. More recently, renewed endeavors have been made to collect the reported cases and to classify them on the basis of histopathology and the more precise knowledge of these neoplasms acquired in the

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1. Of these 313 cases, 261 were supposedly primary in the ribs.

intervening years. Harper, in 1939, collected some 60 cases of benign cartilagenous tumors of the rib. Sommer and Major (1942) reported on 81 cases of primary tumors of the rib—66 gleaned from the literature and another 15 under their personal observation. Six years later Dorner and Marcy reviewed most of the cases of primary tumors of the ribs reported between 1942 and 1947. In order to correlate all this material I have checked the reports of my predecessors, added the cases they omitted, and reviewed 205 cases of primary tumors of the rib, which brings the present study through 1951. Unfortunately, even many of the more recent case reports are incomplete and do not permit definitive conclusions as to etiology, location, and nature of outcome of therapy.

ETIOLOGY

Many theories have been advanced in an attempt to explain the origin of these tumors: (1) hereditary predisposition, (2) presence of supernumerary joint cartilages, (3) maldevelopment, (4) cell nests or displaced cartilaginous rests, (5) endocrine dysfunction, (6) vitamin and nutritional deficiency, (7) infection, and (8) trauma. By far the most commonly discussed theory is that of trauma. Although often invoked as the cause, few, if any, have offered sufficient proof to warrant the acceptance of trauma as the sole possible cause of the tumor. Regarding this subject, Ewing states that

> To establish the relation of trauma several classes of evidence are necessary: (1) the authenticity and sufficient importance of the trauma; (2) previous integrity of the wounded part; (3) a reasonable time relation, from three weeks to three years or more in certain cases; (4) continuity of pathological changes or symptoms in the wounded part and the appearance of the tumor; (5) microscopical proof of the existence of a tumor.
>
> A careful investigation of cases of malignant tumors of supposed traumatic origin generally reveals an absence of one or more of these essential elements especially the previous integrity of the tissues.

There is no doubt that in some of the cases reported all the above postulates were fulfilled. Yet, I personally doubt that one has the absolute right to draw a conclusion of causal relationship except in very rare instances. When one considers the high incidence of injuries to the chest wall during competitive sports like boxing, football, baseball, basketball, hockey, etc., and in military combat, and the low incidence of tumors of the ribs amongst the participants, as well as the low incidence of such tumors in patients who have sustained fractured ribs from any other cause, one is disposed to doubt a causal relationship.

CLASSIFICATION

Although a number of classifications of primary rib tumors have been presented, most of them are incomplete. Therefore I propose the following classification [2] as being reasonably inclusive and complete.

Benign Tumors
- Chondroma
- Osteochondroma
- Myxochondroma
- Osteoma

2. Eosinophilic granuloma of the rib has been omitted because the consensus is that this lesion is inflammatory rather than neoplastic.

Osteoid osteoma
Osteofibroma
Fibroma
Fibrous dysplasia
Hemangioma
Xanthomatous giant cell
Miscellaneous—myxoma, solitary myeloma, xanthoma, lipoma, chondromyxoid fibroma

Malignant Tumors
Chondrosarcoma
Osteogenic sarcoma
Fibrosarcoma
Mixed sarcomas
Endothelioma (Ewing's Tumor)
Hemangiosarcoma
Giant cell
Miscellaneous—myeloma, lymphosarcoma, reticulum-cell sarcoma, hemangioendothelioblastoma, liposarcoma

SYMPTOMS AND SIGNS

The symptom-complex is variable, ranging from complete absence of any manifestations of its presence to a multiplicity of variegated complaints. The symptoms may be subdivided into the following four categories: (1) complete absence, (2) local manifestations, (3) referred symptoms, and (4) systemic manifestations. The symptoms are dependent upon the nature and duration of the local lesion, the rate of growth of the tumor, the extent of involvement of other structures in juxtaposition to the local tumor and/or its distant dissemination, and, finally, its relation to other manifestations of the initial cause.

1. *Absence of Symptoms.*—In such cases the tumor is usually discovered during a "routine" physical examination of the chest or is an incidental roentgenographic finding when the thorax is studied for unrelated conditions. In patients with fibrous dysplasia of the rib, the tumor was asymptomatic in a high percentage of the cases. In one of the cases reported herein by me, an asymptomatic chondroma of the rib was discovered during the preinduction roentgenographic examination of the chest before the patient entered military service.

2. *Local Manifestations.*—By far the two commonest complaints encountered in patients with a primary tumor of the rib are local pain and a mass on the chest wall. The pain is of variable intensity, from that of an intermittent local dull ache to a persistent severe pain in the area of the growth. In benign and/or slowly growing tumors, the pain is more or less of the same intensity over a long period and is often described as dull or boring in nature and unaffected appreciably by movement or deep breathing, whereas in malignant tumors or rapidly expanding growths it is progressive in severity and often worsened by exertion, coughing, straining, and deep breathing. In all cases pleuritic pain implies extension to the pleura.

The tumor may be visible and palpable on the chest wall. In advanced cases it may be accompanied by changes in the overlying skin. It is unpredictable in size, shape, consistency, and degree of fixation within the chest wall. Because of the thickness of the muscles of the back and the position of the scapula, tumors in that area may not be visible or palpable. In other areas the inward growth of the neoplasm may not disclose its presence to the eye or palpating finger. In one of the cases reported herein by me, that of the patient with a fibrosarcoma of the ribs, the com-

TABLE 1.—*Primary Tumors of the Rib*

Case No.	Author	Sex	Age, Yr.	Location of Tumor	Duration of Symptoms	Initial Symptoms	Later Symptoms	Therapy	Comment
Benign Tumors									
Chondroma									
1	Sommer and Major (1942)*	M	11	Rib 2 anterior	.....	Tumor	.....	Partial excision	Well 1 yr. after operation
2	.....	M	26	Rib 10	3 mo.	Mild digestive complaints	Tumor and weight loss	Excision	Survived operation
3	.....	M	3½ mo.	Rib 6 angle	1 mo.	Tumor	.....	Removal with ribs 6 and 7	Survived operation
4	.....	M	37	Ribs 8, 9, and 10 at cartilages	10 yr.	Tumor	Pain	Excision	Survived operation
5	.....	M	38	Ribs 3 and 4 anterior	4 yr.	Tumor	Pain	Removal with ribs 3 and 4	Survived operation
6	.....	F	55	Ribs 3 and 4 anterior	9 yr.	Tumor	Pain	Removal with ribs 3 and 4	Previous incomplete removal necessitated extensive resection later; well 5 mo after last operation
7	.....	F	17	Rib 4 cartilage	1 yr.	Tumor	.....	Removal of tumor and rib	Well 11 yr. after operation
8	.....	M	41	Rib 6 anterior	6 yr.	Pain	.....	Excision	Well 3½ yr. after operation
9	.....	M	30	Rib 3 cartilage	.....	.....	.....	Removal with rib	Survived operation
10	.....	M	47	Rib 7 head and neck	3 mo.	Pain	Tumor	Incomplete removal with ribs 6 to 9	Well 5 yr. after operation
11	Dorner and Marcy (1948) *	F	64	Ribs 1 and 2 anterior	50 yr.	Tumor	.....	Removal	Survived operation
12	Bertola and Ferraris (1946)	F	25	Rib 8	2 yr.	Pain	Tumor and dyspnea	Removal with rib	Survived operation
13	LaVilla (1946)	F	14	Rib 7 posterior	1 mo.	Cough	.....	Removal with rib	Survived operation
14	.....	F	20	Rib 5 posterior	6 mo.	Pain	.....	Removal with rib	Well 1 yr. after operation
15	Cabot Case 32442 (1946)...	F	56	Rib 8 anterior	2 yr.	Pain	Tumor	Removal with rib	.....
16	Aguirre and Martinez (1947)	M	35	Rib 10 anterior	15 days	Tumor	.....	Removal with rib	Survived operation
17	Morton and Mider (1947)	F	45	Ribs 3 and 4 anterior	5 yr.	Tumor	.....	Removal with rib	No local recurrence but chondrosarcoma of ilium developed 12 yr. later and patient died of it at 64 yr. of age
18	Watson and James (1947)	M	18	Rib 4 anterior	2 mo.	.....	.....	Removal with rib	Recurrence in rib stump 4 mo. later requiring extensive resection; survived second operation
19	Dorken (1947)	M	56	Rib 3 anterior	3 yr.	Tumor	.....	Removal	Survived operation
20	Poppe and Berg (1948)....	M	67	Ribs 6, 7, and 8 anterior	5 yr.	Tumor	.....	Removal with ribs	Survived operation
21	Pasquali (1950)	F	32	Rib 2 anterior	Months	Pain	.....	Biopsy	Survived operation
22	Hochberg	M	29	Rib 3 anterior	.....	.....	.....	Removal with rib	No recurrence 6 yr. after operation
23	Hochberg	M	34	Rib 4 anterior	1 yr.	Tumor	Pain	Removal with rib	No recurrence 3 yr. after operation
Osteochondroma									
24	Sommer and Major (1942) *	F	24	Rib 3 anterolateral	1 mo.	Pain	Tumor	Removal with rib	Well 2 yr. after operation
25	.....	M	23	Ribs 7 to 10 anterior	7 yr.	Tumor	Pain and dyspnea	Removal	Postoperative death; necropsy revealed osteochondromata in lung
26	.....	M	21	Rib 1 posterior	4 mo.	Tumor	.....	Removal followed by x-ray	Well 2 yr. after operation
27	.....	M	40	Rib 9 anterior	2 yr.	Pain	Tumor	Removal with rib	Well 11 yr. after operation
28	.....	F	55	Rib 10	1 yr.	Tumor	Pain	Removal with ribs 9, 10, and 11	Well 20 mo. after operation

TABLE 1.—*Primary Tumors of the Rib*—Continued

Case No.	Author	Sex	Age, Yr.	Location of Tumor	Duration of Symptoms	Initial Symptoms	Later Symptoms	Therapy	Comment
29	Dorner and Marcy (1948) *	F	24	Anterior	8 yr.	Tumor	.....	Removal	Well 12 yr. after operation
30	.....	M	51	Rib 1 anterior	6 mo.	Tumor	Pain	Removal with medial part of clavicle	Survived operation
31	.....	F	38	Rib 4 anterior	1 yr.	Tumor	.....	Removal	.....
32	.....	F	33	Rib 2 anterior	15 yr.	Tumor	.....	Removal	Well 16 mo. after operation
33	Janes (1941)	F	16	Anterior chest	3-4 yr.	Tumor	.....	Partial removal	20 yr. later removal of recurrence with local chest wall
34	Mathey and others (1947)	M	12	Ribs 3 and 4	?	Tumor	.....	Removal with ribs	Survived operation
35	Simpson (1948)	F	34	Rib 1 posterior	?	Horner's syndrome	.....	Removal with ribs 1 and 2	Well 8 mo. after operation
36	Poppe and Berg (1948)....	M	65	Ribs 5, 6, and 7 anterior	10 yr.	Tumor	.....	Removal with ribs 5, 6, and 7	Survived operation
37	Hochberg	M	18	Rib 5 anterior	3 yr.	Tumor	.....	Removal with rib	Well 13 yr. after operation
	Myxochondroma								
38	Janes (1941).....	F	65	Anterior	30 yr.	Tumor	.....	None	Autopsy findings
39	Hochberg	M	62	Rib 5 lateral	2 yr.	Tumor	Pain	Removal with rib	Well 1 yr. after operation
	Osteoma								
40	Sommer and Major (1942)*	F	49	Rib 11	4 yr.	Pain	.....	Removal with rib	Survived operation
41	.....	M	42	Rib 5	30 yr.	Tumor	.....	Removal with rib	Survived operation
42	.....	F	26	Rib 6 posterior	2 mo.	Pain	Elevated temperature	Removal with rib	Well 2 yr. after operation
43	Flori (1950).....	M	5	Ribs 3 and 4 posterior	2 mo.	Pain	Anorexia	Removal with ribs	Survived operation
	Osteoid osteoma								
44	Dorner and Marcy (1948)*.	M	11	Rib 9 posterior	8 mo.	Pain	.....	Removal with rib	Well 2½ yr. after operation
45	Hochberg	M	15½	Rib 11 posterior	1 yr.	Pain	Elevation of hip	Removal with rib	Well 3 yr. after operation
	Osteofibroma								
46	Sommer and Major (1942)..	F	28	Rib 1 anterior	18 mo.	Tumor	Pain	Removal with rib	Survived operation
47	Lentino (1950).....	M	22	Rib 4 lateral	8 mo.	Pain	Tumor	Removal with rib	Survived operation
	Fibrous dysplasia								
48	Cabot case 32452 (1946)....	M	27	Rib 2 posterior	Several yr.	Pain	.....	Removal	Survived operation
49	Schlumberger (1946).....	M	27	Rib 1 posterior	3 mo.	Tumor	.....	.....	.....
50	.....	M	29	Rib 2 anterior	16 mo.	.....	.....	.....	.....
51	.....	M	30	Rib 2 lateral	.....	.....	.....	.....	.....
52	.....	M	21	Rib 2 anterior	.....	.....	.....	.....	.....
53	.....	M	31	Rib 3 posterior	.....	.....	.....	.....	.....
54	.....	M	25	Rib 4 anterior	1 yr.	.....	.....	.....	.....
55	.....	M	20	Rib 4 anterior	3½ yr.	.....	.....	.....	.....
56	.....	M	24	Rib 4	8 mo.	.....	.....	.....	.....
57	.....	M	23	Rib 3 posterior	2 yr.	.....	.....	.....	.....
58	.....	M	37	Rib 5 lateral	2½ yr.	.....	.....	.....	.....
59	.....	M	34	Rib 6 lateral	8 yr.	.....	.....	.....	.....
60	.....	M	22	Rib 6 posterior	.....	.....	.....	.....	.....
61	.....	M	22	Rib 5 posterior	.....	.....	.....	.....	.....
62	.....	M	25	Rib 6 posterior	.....	.....	.....	.....	.....
63	.....	M	28	Rib 6 posterior	.....	.....	.....	.....	.....

64	.....	M	19	Rib 6 lateral	.....	.....	.....	Removal	.....
65	.....	M	26	Rib 7 posterior	7 mo.	.....	.....	.....	.....
66	.....	M	23	Rib 7	2 yr.	.....	.....	Removal	.....
67	.....	M	37	Rib 7 lateral	.....	.....	.....	Removal	.....
68	.....	M	22	Rib 7 lateral	1 yr.	.....	.....	.....	.....
69	.....	M	26	Rib 8 posterior	1 yr.	.....	.....	.....	.....
70	.....	M	36	Rib 9 lateral	.....	.....	.....	.....	.....
71	.....	M	33	Rib 9 posterior	4 mo.	.....	.....	.....	.....
72	.....	M	30	Rib 9 posterior	2½ yr.	.....	.....	.....	.....
73	.....	F	37	Rib 9 posterior	.....	.....	.....	.....	.....
74	.....	M	29	Rib 10 anterior	8 yr.	.....	.....	.....	.....
75	.....	M	30	Rib 10 anterior	6 yr.	.....	.....	.....	.....
76	.....	M	27	Rib 10 lateral	8 mo.	.....	.....	.....	.....
77	.....	M	34	Rib 11 posterior	.....	.....	.....	.....	.....
78	Dorner and Marcy (1948)..	F	21	Rib 6 posterior	.....	.....	.....	Removal	Well 10 mo. after operation
79	Vilvandre (1950).....	M	45	Rib 4 anterior	.....	.....	.....	Removal	.....
80	Flickinger (1951).....	M	41	Ribs 3, 4, and 5 anterior	7 mo.	Pain	Tumor	Removal with ribs	Well 3 yr. after operation
81	Lichtenstein and Jaffe (1942)	F	35	Several ribs	.....	.....	.....	.....	.....
82	Hochberg	M	37	Ribs 6, 7, and 8 posterior	1 mo.	Pain	.....	Removal with ribs	Well 5 yr. after operation
	Hemangioma								
83	Urteaga Ballon, Peschiera, and Bernal (1947).....	M	23	Rib 9 posterior	2 yr.	Cough	Pain and tumor	Removal	Survived operation
84	Aksoy and Güchan (1947).	M	42	Rib 7 lateral	2½ yr.	Tumor	Pain	Removal	Survived operation
85	Dorner and Marcy (1948)*.	M	34	Rib 6 posterior	15 yr.	Pain	.....	Removal	Survived operation
86	.....	F	22	Rib 9 posterior	8 yr.	Tumor	Pain	Removal with rib	Well 8 mo. after operation
87	Gauwerky and Hartjen (1951)	M	45	Rib 6 lateral	3 yr.	Pain	Dyspnea	Removal	Survived operation
88	.....	F	54	Rib 6 posterior	6 yr.	Pain	Cough and expectoration	Removal with ribs 5, 6, and 7	Survived operation
89	.....	F	61	Anterior	9 mo.	Tumor	Pain	Removal	Case reported by Oehlecker in 1948
90	Hochberg	F	31	Rib 3 posterior	2 yr.	Pain	.....	Removal with rib	Well 1 yr. after operation
	Xanthomatous giant cell								
91	Sommer and Major (1942)*.	M	11	Rib 1 and clavicle	8 mo.	Tumor	.....	Removal with rib	Well 17 yr. after operation
92	.....	F	14	Rib 12	1 yr.	Pain	Tumor	Removal and radium	Well 13 yr. after operation
93	.....	M	17	Rib 10 posterior	9 mo.	Tumor	Weight loss	X-ray and removal with ribs 8 to 12	Benign giant-cell tumor with chondromatous and osteogenic sarcoma; postoperative death
94	.....	F	38	Rib 8 posterior	3 yr.	Pain	.....	Intercostal neurectomy for pain	Well 11 yr. after operation
95	.....	M	48	Rib 5 anterior	6 mo.	Tumor	.....	Removal	Well 11 yr. after operation
96	.....	M	9	Rib 4 lateral	2 mo.	Pain	Tumor	Biopsy and radium	Well 11 yr. after operation
97	.....	F	31	Rib 2 anterior	2 yr.	Tumor	.....	Removal	Well over 5 yr. after operation
98	.....	F	15	Rib 5 lateral	6 mo.	Tumor and pain	.....	Biopsy and x-ray	Well 2 yr. after operation
99	.....	M	47	Rib 7 lateral	10 mo.	Pain	.....	Removal	Died 2½ yr. after operation of carcinoma of lung
100	Buckles and Lawless (1950)	M	49	Rib 5 anterior	8 yr.	Tumor	Pain, malaise, weakness, fever, ulceration of skin	Biopsy, x-ray	Well 5 mo. after operation

TABLE 1.—*Primary Tumors of the Rib*—Continued

Case No.	Author	Sex	Age, Yr.	Location of Tumor	Duration of Symptoms	Initial Symptoms	Later Symptoms	Therapy	Comment
	Miscellaneous								
101	Sommer and Major (1942)*.	M	27	Rib 10 posterior	Months	Pain	.....	Removal	Fibroma; survived operation
102	Marottoli (1946).....	M	20	Rib 9 lateral	1 mo.	Pain	Tumor	Removal and x-ray	Myeloplaxes; well 6 yr. after operation
103	Aufses (1948).....	M	58	Rib 3 lateral	6 mo.	Pain	.....	Removal with ribs 2, 3, and 4	Myeloma; died 3 yr. after operation of coronary artery occlusion; no evidence of recurrence
104	Prevost (1950).....	M	36	Rib 8 posterior	.....	Pain	.....	Removal	Xanthoma; survived operation
105	Fesani (1951).....	M	76	Rib 2 anterior	15 mo.	Pain	Tumor	Removal with ribs 2 and 3	Myxoma; recurred twice and mass removed each time
106	Goorwitch (1951).....	M	39	Rib 9 posterior	7 yr.	Pain	.....	Removal with rib	Chondromyxoid fibroma; survived operation
	Malignant Tumors								
	Chondrosarcoma								
107	Sommer and Major (1942)*.	M	29	Ribs 8, 9, and 10 lateral	4 yr.	Tumor	.....	Removal with ribs	Well 3 yr. after operation
108	.....	F	42	Several ribs	3½ yr.	Tumor	.....	Partial removal	Postoperative death
109	.....	M	38	Several ribs	2 yr.	Tumor	Pain	Partial removal	Died of metastasis 3 yr. after operation
110	.....	M	41	Rib 12	.....	.....	.....	Removal	Recurred 8 mo. after operation
111	.....	M	62	Ribs 2, 3, and 4 anterior	3 yr.	Tumor	.....	Removal with ribs	Recurred 6 mo. after operation
112	.....	M	40	Ribs 2, 3, and 4 anterior	5 yr.	Tumor	Pain	Removal	Died 6 yr. after operation of recurrence
113	.....	M	34	Ribs 8, 9, and 10 posterior	8 mo.	Tumor and pain	.....	X-ray and partial removal	Died 7 mo. after operation
114	.....	F	23	Ribs 5 and 6 lateral	6 mo.	Pain	.....	Removal with ribs	Died 25 mo. after operation
115	Dorner and Marcy (1948)*.	F	60	Ribs 7 and 8 anterior	10 yr.	Pain	Tumor and dyspnea	Removal	Recurred 6 yr. after operation; second removal with recurrence 3 yr. later
116	.....	F	47	Rib 3 anterior	3 yr.	Tumor	Pain	Removal with ribs	No recurrence over 5 yr. after operation
117	.....	M	39	Rib 4 posterior	2 yr.	Pain and tumor	.....	X-ray and removal	Survived operation
118	.....	M	16	Rib 10 posterior	3 days	Cough	Pain and dyspnea	Biopsy	Died 1 mo. after operation
119	.....	M	45	Ribs 9 and 10 anterior	15 yr.	Tumor	.....	Partial removal	.....
120	.....	F	51	Rib 11 lateral	4 yr.	Pain	Tumor	Removal	Died 1 yr. after operation
121	.....	M	56	Ribs 6 and 7 lateral	3 yr.	Tumor	Pain	Partial removal	Died 3 mo. after operation
122	Janes (1941).....	M	63	Rib 3 anterior	1 mo.	Tumor	.....	Removal and x-ray	Died 1 yr. after operation
123	.....	M	60	Ribs 9, 10, and 11 anterior	20 mo.	Tumor	.....	Removal with ribs	Well 1 yr. after operation
124	Jaffe (1943).....	M	?	Rib 6 lateral	2 yr.	Tumor	.....	Partial removal	Recurrence 4 mo. after operation; 2d operation with recurrence again
125	Pendergrass and others (1945)	M	55	Rib 7 lateral	1 yr.	Tumor	.....	Removal with rib	Well 7 yr. after operation
126	.....	F	39	Rib 11 posterior	1 yr.	.....	.....	Removal with rib	Well 6 yr. after operation
127	Ducuing and Marquès (1945)	F	38	Posterior	4 yr.	Pain	Tumor	Biopsy	.....
128	Morton and Mider (1947)..	F	27	Ribs 7, 8, and 9	1 yr.	Tumor	.....	Removal	Recurred 15 mo. after operation; removed and recurred again; died 6 yr. after first operation

129	Watson and James (1947).	F	53	Ribs 9 and 10 anterior	2 yr.	Tumor	.....	Removal with ribs 9 to 12	Died 18 days after operation of pulmonary embolism
130	Davis, Tollman, and Brush (1949)	F	25	Ribs 5, 6, and 7 anterior	2 yr.	Tumor	Dyspnea	Removal with ribs	No recurrence 3 mo. after operation
131	Condon and Harper (1950)	F	60	Lower thorax and upper abdomen anterior	13 yr.	Dyspnea and pain	Tumor	Removal *in toto*	Recurrence years after operation requiring 2d operation; alive 10 yr. after first operation
132	O'Neal and Ackerman (1951)	M	45	Rib 5 anterior	7 mo.	Pain	.....	Removal with ribs	Recurred 2 mo. after operation; removed with surrounding tissues; died 2+ yr. after first operation
133	.....	F	68	Rib 4 anterior	8 mo.	Tumor	.....	Removal	Recurred several times; reoperation after each recurrence; died 4 yr. after first operation
134	.....	M	61	Rib 3 anterior	3 yr.	Tumor	.....	Removal with ribs 2 to 4	Recurred 1 yr. after operation, partial resection performed; died 2 yr. after first operation
135	.....	M	36	Rib 12 posterior	3½ mo.	Pain	.....	Removal with ribs 10 to 12	Recurred 6 mo. after operation; died 11 mo. after operation
136	.....	F	25	Rib 7 posterior	6 mo.	Pain	.....	Partial removal	.....
137	.....	M	74	Rib 9 anterior	?	Tumor	.....	Biopsy	Died 22 mo. after onset of symptoms
138	.....	M	60	Rib 10 anterior	6 mo.	Tumor	.....	Removal with ribs	Well 18 mo. after operation
139	.....	M	70	Rib 3 posterior	3 mo.	Pain	.....	Partial removal	Survived operation
140	.....	M	58	Rib 9 anterior	6 yr.	Tumor	.....	Removal with ribs 8 to 10	Well 3+ yr. after operation
141	Guglielmini and Pironti (1951)	M	30	Rib 7	1 yr.	Pain	Tumor	X-ray and partial removal	Died 6 mo. after operation
142	Hochberg	M	32	Ribs 6, 7, and 8 posterior	4 yr.	Pain	Tumor	Partial removal	Originally operated on elsewhere; admitted 8 mo. after operation; died soon after entering hospital
	Osteogenic sarcoma								
143	Sommer and Major (1942)*	M	44	Rib 10	1 wk.	Tumor	.....	Removal	Well 3 yr. after operation
144	.....	F	27	Rib 7	9 mo.	Pain	Tumor	Removal with ribs	Well 18 mo. after operation
145	.....	M	32	Ribs 2 to 5 anterior	1 yr.	Tumor	Dyspnea and palpitation	Removal with ribs and adjacent lung tissue	Survived operation
146	.....	M	54	Rib 11 posterior	1 wk.	Deformity of chest wall	Tumor	Removal with ribs 10 and 11	Well 1 yr. after operation
147	Lugue (1944).....	M	?	Rib 7 posterior	2 yr.	Tumor	Epigastric pain	Removal with rib	Well 2 mo. after operation
148	Johnson (1951).....	M	27	Rib 8	?	Pain	.....	Removal	Recurrence 1 yr. after operation; reoperation 6 yr. later for mediastinal mass, which was asymptomatic 2 yr. later
	Fibrosarcoma								
149	Sommer and Major (1942)*	M	61	Rib 3 anterior	1 yr.	Tumor	.....	Removal with ribs 2, 3, and 4	Died some months after operation
150	.....	M	41	Ribs 8 and 9 lateral	6 mo.	Tumor and pain	.....	Biopsy and x-ray	Living 10 mo. after operation
151	.....	F	34	Axillary part of chest	2 yr.	Tumor	.....	Removal with 4 ribs	Died 1 yr. after operation of metastasis
152	.....	M	42	Ribs 1 and 2 anterior	4½ yr.	Tumor	Pain	Removal with clavicle	Previously removed and received x-ray therapy; survived 2d operation
153	.....	F	46	Ribs 7, 8, and 9 posterior	2 yr.	Pain	Tumor	Biopsy and x-ray	Died 2 mo. after operation

TABLE 1.—*Primary Tumors of the Rib*—Continued

Case No.	Author	Sex	Age, Yr.	Location of Tumor	Duration of Symptoms	Initial Symptoms	Later Symptoms	Therapy	Comment
154	.....	F	50	Scapular area	4 mo.	Pain	Tumor	Removal with ribs 2 to 8	Died after operation
155	Blake and Bradford (1943)	M	20	Rib 6 lateral	5 mo.	Pain	Tumor and anorexia	X-ray; neurectomy for pain	Died 21 mo. after onset of symptoms
156	Dorner and Marcy (1948)..	M	37	Ribs 9 and 10 anterior	6 mo.	Pain	Tumor	Removal	Died 9 mo. after operation
157	Blades and Paul (1950)...	M	25	Rib 4 posterior	2 yr.	Malaise	Elevation of temperature and pain	Removal	Died of recurrence 1 yr. after operation
158	.....	F	19	Rib 7 lateral	3 wk.	Tumor	.....	Removal with rib	Well 5 yr. after operation
159	Guglielmini and Pironti (1951)	M	10	Subscapular area	3 yr.	Pain	Tumor	Removal with ribs 7 to 10	Died of recurrence 2 mo. after operation
160	Hochberg	F	36	Rib 5 posterior	2 yr.	Pain	Tumor	Removal with ribs 5 to 8	Recurred 3 times, twice removed but not third time because of vertebral invasion; alive but not well 4+ yr. after onset of symptoms
	Mixed sarcomas								
161	Sommer and Major (1942)*	F	12	Rib 7	?	Path. fracture	.....	Removal	Osteogenic myxofibrosarcoma; survived operation
162	.....	F	43	Ribs 4 and 5 anterior	?	Pain	Tumor	Removal with ribs	Chondromyxosarcoma; well 14 mo. after operation
163	.....	M	66	Interscapular area	8 yr.	Tumor	Pain	Partial removal	Osteogenic myxochondrosarcoma; died 2 mo. after operation
164	.....	F	38	Anterior	2 yr.	Tumor	.....	Removal and x-ray	Chondroosteosarcoma; recurred 6 mo. after operation
165	Dorner and Marcy (1948)*	M	33	Rib 10	1 yr.	Pain	.....	Removal and x-ray	Osteochondromyxosarcoma; well 4 mo. after operation
166	Lentino (1950).....	M	?	Rib 9 posterior	20 yr.	Tumor	.....	Removal and x-ray	Osteochondrosarcoma; recurred; given x-ray therapy and 11 yr. later removed again; alive 13 yr. after first operation
167	.....	M	32	Rib 6 anterior	4 mo.	Tumor	.....	Removal	Fibrochondrosarcoma recurred twice, requiring extensive resection each time; died 3 yr. after first operation
168	LeBihan and Boiset (1951)	M	23	Rib 11 posterior	18 mo.	Tumor	Pain and weight loss	Removal with ribs 10 to 12	Fibromyxomatous osteosarcoma; recurrence 4 yr. later; requiring removal; recurrence 4 mo. later; given x-ray therapy
	Ewing's tumor								
169	Sommer and Major (1942)*	M	15	Rib 4 lateral	9 mo.	Pain	Tumor	Removal with ribs 3, 4, and 5	Died of recurrence 5 mo. after operation
170	.....	F	15	Ribs 4 to 8 posterior	5 mo.	Pain	Tumor	Partial removal	Died 3 mo. after operation; metastasis
171	.....	F	18	Rib 9 posterior	3 mo.	Pain	Pleurisy	Partial removal	Died 10 mo. after operation; metastasis
172	.....	F	15	Rib 4 posterior	18 mo.	Pain and tumor	.....	Removal	Recurred 18 mo. later; mass removed; died a few months later
173	.....	M	27	Ribs	3 mo.	Dyspnea	Tumor	X-ray	Died
174	.....	M	10	Rib	4 mo.	Pain	.....	X-ray	Well 2 mo. later
175	.....	M	12	Ribs 11 and 12 and 11th and 12th thoracic vertebrae	6 mo.	Pain	Tumor; paraplegia	Biopsy and x-ray	Died 6 mo. later

176	.....	M	16	Rib 11	3 mo.	Tumor	Pain	Removal	Recurred 3 times; mass removed each time; died of recurrence
177	.....	M	10	Rib 6 lateral	3 mo.	Pain	Fever; pleurisy	Removal and x-ray	Died 1 mo. later
178	.....	M	20	Rib 3 lateral	3 mo.	Pain; tumor	Loss of weight	Biopsy and x-ray	Died soon thereafter
179	.....	M	15	Ribs 5 and 6 lateral	15 days	Pain	Tumor	Removal and x-ray	Died 6 mo. later; metastasis
180	.....	M	10	Ribs 6 and 7 lateral	6 mo.	Malaise	Tumor and pain	Partial removal	Alive 18 mo. after operation
181	.....	M	10	Ribs 6 and 7 lateral	1 mo.	Tumor	Pain	X-ray and removal	Survived operation
182	.....	M	14	Rib 8 posterior	12 mo.	Pain	Tumor and dyspnea	X-ray	Survived operation
183	.....	M	22	Rib	12 mo.	Pain	.....	Curettage	Died 3 yr. after operation
184	.....	M	30	Rib	6 mo.	Cough and pleurisy	Pain	X-ray	Died 2 yr. after operation
185	.....	M	11	Rib	3 mo.	Pain	Pleural effusion	X-ray	Died 3 mo. after operation
186	.....	F	22	Rib 11 posterior	2 mo.	Pain	Tumor	Biopsy and x-ray	Died 7 mo. after operation
187	Cabot Case 32091 (1946)...	M	18	Rib 1 anterior	4 mo.	Pain	.....	Removal	Survived operation
188	.....	M	5	Rib 8	.....	.....	.....	Biopsy	.....
189	Sgrosso and Fontana (1946)	M	31	Rib 11 posterior	6 mo.	Pain	Tumor	Removal with rib	Recurrence 5 mo. after operation
190	Cabot Case 33401 (1947)...	F	30	Rib 7 anterior	18 mo.	Tumor	Dyspnea	Removal	.....
191	Kent and Ashburn (1948)..	M	19	Rib 5 lateral	12 mo.	Tumor	Pain	X-ray; removal with ribs 4 to 6 and involved lung tissue	Well 13 mo. after operation
192	Hochberg	M	20	Rib 11 posterior	2 mo.	Pain	.....	Removal of ribs 11 and 12	No recurrence 1 yr. after operation
	Malignant giant-cell tumor								
193	Guglielmini and Pironti (1951)	F	59	Rib 3	Months	Tumor	Pain	Removal and x-ray	Died 3 mo. after operation
	Miscellaneous malignant tumors								
194	Sommer and Major (1942)*	M	23	Rib 12 posterior	3 mo.	Tumor	.....	Removal with rib	Round-cell sarcoma; recurred 2 mo. later and removed again
195	.....	F	29	Rib 12 posterior	6 mo.	Tumor	.....	Removal	Small round-cell sarcoma; postoperative death
196	.....	M	4	Ribs 3, 4, and 5 anterior	4 mo.	Tumor	.....	Removal with right middle lobe	Round-cell sarcoma; survived operation
197	.....	F	26	Rib 10 posterior	1+ yr.	Pain	Tumor	Removal	Round-cell sarcoma; survived operation
198	.....	M	9	Rib 10	5 yr.	Pain	Tumor	Biopsy and x-ray	Lymphosarcoma; died 6 mo. after operation
199	.....	M	26	Ribs 9, 10, and 11	6 yr.	Pain	Tumor	Removal	Lymphosarcoma; survived operation
200	Dorner and Marcy (1948)*	M	60	Ribs 4 to 7 lateral	3 mo.	Pain	Weight loss, headache, and chills	X-ray	Plasmocytoma; well 5 mo. after onset
201	.....	F	27	Rib 9 anterior	2 yr.	Pain	Tumor; weight loss	Removal	Lymphocytoma; no recurrence 3 yr. after operation
202	Holden (1949).....	F	60	Rib 1	6 mo.	Pain and tumor	.....	Biopsy and x-ray	Plasmocytoma; well 10 mo. after onset
203	Coley and others (1950)...	M	63	Rib 8	.....	.....	.....	X-ray	Reticulum-cell sarcoma; well 6½ yr. later
204	.....	M	57	Rib 12 posterior	.....	.....	.....	Removal	Reticulum-cell sarcoma; recurrence 11 mo. later; removed again; Coley toxin and x-ray; well 2 yr. after first operation
205	Hochberg	M	49	Ribs 5, 6, and 7 lateral	3 wk.	Tumor	.....	Removal with ribs	Hemangioendothelioblastoma; alive 1 yr. after operation

* Collected cases.

plaints were severe local pain in the region of a progressively enlarging tumor in the axillary area. In another case, that of the patient with a hemangioma of the rib, there was severe local pain without any external manifestations of a tumor.

3. *Referred Symptoms.*—By impingement upon various nerves in proximity of the tumor, corresponding referred symptoms may be evoked. Involvement of the cervical sympathetic nerves will produce symptoms referable to the neck, face, and eye, as in the case reported by Simpson; by involvement of some of the components of the brachial plexus, pain will be elicited in the neck, shoulder, and arm, as in the case reported by Aufses; by involvement of the lower intercostal nerves it will give rise to abdominal symptoms, as in one of the cases detailed in this report, and by involvement of the lumbar plexus it will induce pain in the small of the back, radiating to the pelvic area, as in another case reported in this paper.

4. *Systemic Manifestations.*—Such symptoms and signs are the result of (*a*) a local expression of a systemic disease, (*b*) a malignant primary tumor with extension to other parts of the body, and (*c*) a benign tumor located in such position or growth in size as to encroach upon and interfere with the function of other structures. While the initial symptoms in most cases of neoplasm of the rib are usually pain and tumefaction, in these cases it is not uncommon to encounter also such symptoms as dyspnea, a pressure sensation in the chest, palpitation, anorexia, fever, and loss of weight.

The diagnosis of a primary neoplasm of the rib is often one of exclusion; i. e., it excludes the lesion as a metastatic deposit or as a local expression of a systemic disease. When these are ruled out as the source of the tumor in the rib, the diagnosis still remains inconclusive without adequate roentgenographic studies of the chest wall. In addition to the conventional posteroanterior, lateral, and Bucky roentgenograms, investigation may require tomographic studies and also x-ray studies of the chest after the induction of pneumothorax to help delineate the position of the tumor with respect to the neighboring structures. Many of the roentgenographic characteristics of the tumor depend upon its location, size, manner of growth, and specific histopathological characteristics. Some points of their roentgenographic appearance will be presented when the respective tumors are discussed.

INCIDENCE

There were 205 cases of primary rib tumors, of which 106, or 51.8%, were benign and 99, or 48.2%, malignant.

BENIGN TUMORS

Chondroma.—There were 23 cases of primary chondroma of the ribs, comprising 21.7% of the benign tumors and 11.2% of all the rib tumors. Ten of these tumors occurred in female and 13 in male patients. The ages of the patients ranged from 3½ months to 67 years, with about half the patients being between 25 and 45 years of age. The duration of symptoms varied from 2 weeks to 50 years. Excluding the cases in which symptoms were of short or of very long duration, the average duration of symptoms was between two and three years. The most frequently encountered symptoms were tumor, in 13 cases (in 12 of which it was the initial symptom), and pain, in 10 cases (in 6 of which it was given as the first symptom). In three cases the chondroma was asymptomatic.

While the physical examination may suggest the presence of a tumor of the rib, roentgenographic examination is essential for a more definitive diagnosis. Roentgenographically, chondromas usually appear as a local well-demarcated expansion of a rib with a more or less uniform circumscribed density often simulating a cystic area. The periphery of the opacity often has a knurled appearance. In the 21 chondromas in which the location of the tumor is recorded the anterior or cartilaginous portion of the rib was involved in 17, or 81.6%, of the cases.

These tumors are slow growing and noninvasive. They vary in size from that of a "small nut" to that of a "massive knobby tumor the size of a child's head." While in most instances they are confined to a single rib, not infrequently they grow large enough to involve several ribs, as in the case reported by Sokolov (cited by Sommer

TABLE 2.—*Distribution of Tumors*

	No. of Cases	Percentage of Benign Tumors	Percentage of All Tumors
Benign tumors	106		51.8
Chondroma	23	21.7	11.2
Osteochondroma	14	13.2	6.8
Myxochondroma	2	1.9	1.0
Osteoma	4	3.8	1.9
Osteoid osteoma	2	1.9	1.0
Osteofibroma	2	1.9	1.0
Fibrous dysplasia	35	33.0	17.0
Hemangioma	8	7.6	3.9
Xanthomatous giant cell	10	9.4	4.9
Miscellaneous (myxoma, solitary myeloma, xanthoma, lipoma, fibroma, chondromyxoid fibroma)	6	5.7	2.9
		Percentage of Malignant Tumors	
Malignant tumors	99		48.2
Chondrosarcoma	36	36.3	17.5
Osteogenic sarcoma	6	6.0	2.9
Fibrosarcoma	12	12.1	5.8
Mixed sarcomas	8	8.0	3.9
Endothelioma (Ewing's tumor)	24	24.2	11.7
Giant cell	1	1.0	0.5
Miscellaneous (myeloma, lymphosarcoma, reticulum cell, hemangioendothelioblastoma, hemangiosarcoma, liposarcoma)	12	12.1	5.8

and Major) and that reported by Poppe and Berg. These tumors are grey-pink in color and somewhat elastic in consistency. On the cut surface they are of more or less uniform cartilaginous appearance. Histologically these tumors are comprised of well-differentiated, mature, and uniform cartilage, but they are not as well formed as normal cartilage and therefore not as completely hyaline in composition.

Complete excision is usually not followed by recurrence. Watson and James, however, report a case in which complete removal was followed by local recurrence at the stump of the resected rib four months after its removal. Morton and Mider report the case of a 45-year-old woman in whom complete removal of the rib tumor was followed 12 years later by a chondrosarcoma of the ilium. She died of that condition at the age of 64 years. At no time were there any signs of recurrence of the tumor of the chest wall.

A 29-year-old man entered the hospital on March 12, 1945, for the removal of an asymptomatic tumor of the rib. He was rejected for military service because preinduction roentgenographic examination of the chest showed the presence of a tumor on the anterior part of the left third rib. At no time did the patient have any symptoms referable to that area. Physical

examination revealed the presence of a discrete, firm, nodular area within the substance of the anterior part of the left third rib. Roentgenographic examination of the chest (Fig. 1*A*) showed a circumscribed mass in the cartilaginous portion of the left third rib measuring 2 by 1.5 cm. Although the lesion was within the rib proper, it extended beyond the linear delineation of the normal rib contour.

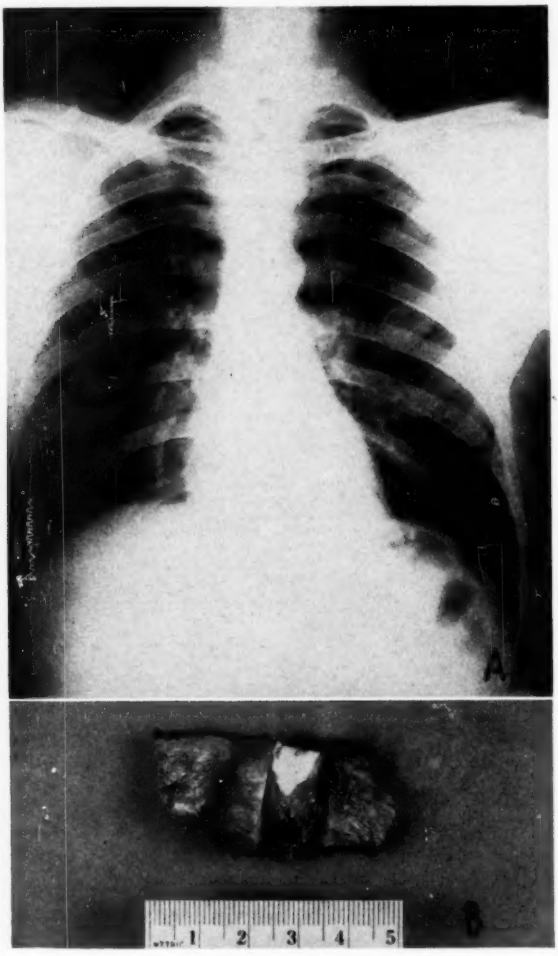

Fig. 1.—*A*, roentgenogram of the chest showing an expanded cyst-like tumor in the anterior part of the third rib on the left side.

B, extirpated tumor (S. P. 45-785) shown in *A*. It consists of a firm nodular tumor, cut across in its bulbous area and reposited between the cut ends of the neoplasm in the rib.

On March 14, 1945, the tumor with surrounding normal rib was removed.

The pathologist's report of the rib (Fig. 1*B*) was as follows: The specimen consists of a flat portion of osseous tissue measuring 5.2 by 2.5 by 0.7 cm. removed from the anterior portion of the left third rib. In its midportion it is bulbous and measures 2.6 by 2.2 cm. The external surface of the bulbous portion is fairly firm and nodular. The cut surface shows osseous tissue at one end. The greater portion of the cut surface is homogeneously grey-white with a central

yellow-grey discoloration. Microscopy shows thickened periosteum at one end and beyond it, bundles of striated muscle. The bony trabeculae are delicate or broad and contain narrow spaces composed of adipose tissue with the usual blood elements. The tumor itself consists of a matrix of pink and lavender staining homogenous tissue in which there are embedded pleomorphic cartilage cells of varying size and shape. In places at the junction of tumor and bone the bony spicules have ragged edges. The diagnosis was chondroma of the rib.

Osteochondroma.—There were 14 cases of osteochondroma of the rib, constituting 13.2% of the benign tumors and 6.8% of all the rib tumors. The ages of the patients ranged between 12 and 65 years, with equal distribution between the sexes. The duration of symptoms varied from 1 month to 15 years, with about half the patients having symptoms referable to the tumor for 1 to 3 years. The most frequent symptoms were tumor, in 13 cases, of which it was the initial complaint 11 times, and pain, in 4 cases, of which it was the initial symptom twice.

The roentgenographic findings are essentially the same as those observed in chondromas of the rib. However, mottled areas of calcification may be noted within the tumor and along its periphery. In the 12 cases in which the tumor was localized roentgenographically or at the time of surgical intervention, it was confined to the anterior portion of the rib in 10 cases and to the posterior portion in 2 instances.

To all intent and purposes these tumors have all the gross and histopathologic characteristics of chondromas except for scattered areas of bony spicules within the substance of the tumor. These tumors, too, are usually confined to a single rib. However, Poppe and Berg, Mathey, and Auchincloss report cases in which more than one rib was involved in the neoplastic process. Auchincloss reported the case of a 23-year-old man in whom the 7th to the 10th ribs, inclusive, were involved in the tumor. The patient died soon after the removal of the tumor of the chest wall. Microscopy showed the tumor of the rib to be an osteochondroma. Necropsy revealed similar osteochondromas in the lung. Although this case was originally reported as an osteochondroma, there is some question as to whether the lesions in the lung were not metastatic deposits from a primary sarcoma of the rib.

An 18-year-old man entered the hospital on March 7, 1938, because of a tumor on the right side of the chest. It had been present for about three years but began increasing in size and sensitivity a few months before admission to the hospital. Physical examination revealed a tumor mass on the right anterior chest wall, about 2 in. (5 cm.) in diameter and fixed to the fifth rib. Roentgenographic examination of the chest revealed a localized tumor of the fifth anterior rib on the right side, the center of which was translucent and the periphery of which contained dense bony tissue (Fig. 2*A*).

On March 9 the neoplasm with neighboring normal cartilage and bone was removed.

The pathologist's report of the tissue removed was as follows: The specimen (Fig. 2*B*) is an ovoid shaped mass of firm tissue measuring 7 by 2.5 by 2.5 cm. The middle portion of the mass is grey, coarsely granular, and composed of cartilage-like material. Most of the cut surface is translucent blue-white and well demarcated from the surrounding red spongy osseous tissue. The microscopy shows cartilaginous plates with central portions exhibiting osseous changes. In other areas the tumor is invaded by bony trabeculae. These trabeculae contain well-formed haversian systems.

A diagnosis of osteochondroma was made.

Myxochondroma.—There were two cases of myxochondromas in this series—one reported by Janes and one presented by me. These tumors, too, have many of the clinical and roentgenographic appearances of the chondroma of the rib. However, areas of lesser translucency and transparency are notable within the tumor. On cut section these tumors show cartilaginous overgrowth with foci of gelatinous changes. While the myxochondroma may be the result of nutritional myxomatous

degeneration of a chondroma and therefore should be classified as a chondroma with such degeneration, it is placed here as an entity in order to emphasize the fact that myxomatous degeneration in an otherwise benign tumor does not imply malignant changes.

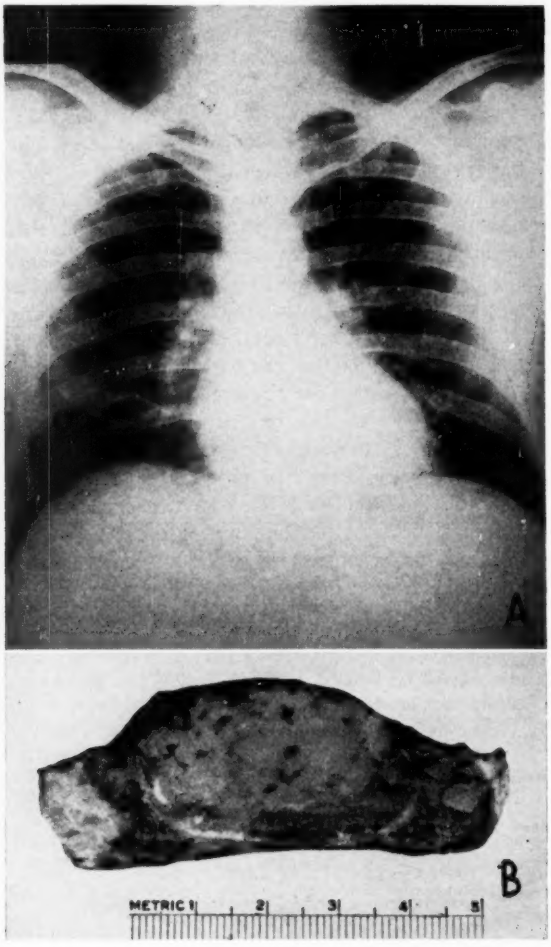

Fig. 2.—*A*, roentgenogram of the chest, revealing a tumor of the fifth rib anteriorly on the right side. Margins of the tumor are of greater density than the body of the neoplasm. Within the substance of the expanded area there are some areas of increased density.

B, tumor (S. P. 38-718) removed from the patient whose roentgenogram is shown in *A*. Note areas of calcification within the tumor and at its periphery.

A 62-year-old man entered the hospital on Jan. 3, 1949, complaining of a mass on the left side of the chest which had been increasing in size for the previous two years. During this period it had given rise to increasing pain locally and over the greater part of the left hemithorax. Physical examination revealed a lobulated firm mass over the left side of the chest, in the anterior axillary area, which was either part of the underlying fourth and fifth ribs or fixed to them.

Roentgenographic examination of the chest (Fig. 3*A*) showed an expanded lesion on the left side of the chest, involving the major portion of the lateral aspect of the fifth rib. The borders of the mass were sclerotic and well defined with no evidence of periosteal reaction in that area. The findings were consistent with those of a benign tumor of the rib.

On Jan. 25 the local tumor was removed together with normal rib in both directions of its long axis.

The pathologist's report of the rib (Fig. 3*B*) was as follows: The specimen consists of a rib measuring 18 cm. in length and presenting in its midportion a large fusiform swelling with many translucent and opaque cystic areas roofed over by thin shell-like bone. This swelling measures 6 cm. in width and 10 cm. in length. On cut section the specimen showed epithelial-lined cystic spaces, the largest of which measures 2 cm. in diameter. The remainder of the cavity is filled with a soft alveolar type of tissue, which is spongy and fibrillar in character. There are several hemorrhagic areas present, and one of the cysts has a small amount of clotted blood in its lumen. Microscopy shows cystic spongy areas lined by flat cells. The spaces contain mucoid material on the outside of which there are fibrillary deposits in a predominantly hyaline cartilage matrix.

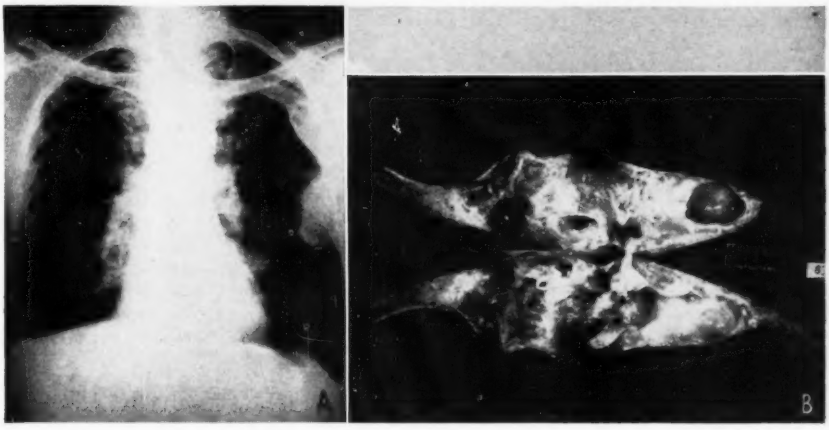

Fig. 3.—*A*, there is a large lobulated tumor present on the anterolateral part of the fifth rib on the left side.

B, rib and tumor removed from the patient whose roentgenogram is shown in *A*. Tumor is fusiform in shape and contains several cystic areas filled with dark blood and light mucoid material.

The tumor was diagnosed as myxochondroma.

Osteoma.—These tumors occurred in 4 cases and comprised 1.9% of the 205 primary rib tumors and 3.8% of the 106 benign tumors. The initial symptoms were pain in three cases and the presence of a growth on the chest wall in the fourth case. The roentgenographic examination of the involved area showed an irregularly expanded lesion with considerable sclerotic change in the neoplastic process. In only one instance, the case reported by Flori, was more than one rib involved. From the descriptions in the literature, it appears that these tumors are hard greyish masses with irregular and firm surfaces. The cut surfaces present bony changes similar to those seen in other osseous tissues. Histologically these tumors are composed of fairly mature bone cells in irregular nests of various phases of growth which are less uniformly organized than mature bone.

Osteoid Osteoma.—There is but one case of an osteoid osteoma of a rib reported in the literature, i. e., that of Ponseti and Barta (cited by Dorner and Marcy). I am reporting another case. These tumors, regardless of their location, are characterized clinically by pain—occasionally intermittent, but always progressive in severity. Roentgenographic examination of the rib or of any other bone with an osteoid osteoma is completely insignificant in the earliest stages. However, as time passes, characteristic changes take place—periosteal thickening takes place in the region of the tumor and at the same time there develops a small focal radiolucent area in the bone which becomes surrounded by osteosclerotic tissue. Grossly, these tumors do not differ from that of a vascular osteoma except that they are usually small growths. The microscopy of these tumors is distinct. The area surrounding the central body is rather sclerotic. Within the central nidus there are many mesenchymal cells, with prominence of preosteal elements, osteogenic connective tissue and irregular areas of calcific masses or trabeculae. All of this is embedded in a highly fibrovascular tissue. Within the substance of the tumor process there may be noted multinucleated osteoplastic giant cells.

A 16-year-old boy was seen by his physician for the first time on Nov. 11, 1949, because of pain in the right flank of about a year's duration. The pain was most disturbing at night. The pain became worse on movement, exertion, and coughing. About six months before entering the hospital, the patient noticed that he favored the right hip, that that side of the pelvis had become elevated, and that there was a prominence of the left side of the back. Physical examination disclosed an area of tenderness in the right inferior costovertebral area and a zone of distinct tenderness to light pressure opposite the right 11th rib in the same area. The right iliac crest and scapula were unduly prominent. There was prominence of the left lumbar musculature, most pronounced on bending. There was also present a left lumbodorsal scoliosis with the concavity to the right. The patient walked with a limp, favoring the right hip. An x-ray of the local area (Fig. 4*A*) revealed a scoliosis of the lumbodorsal area with an area of radiolucency in the medial portion of the 11th rib in close proximity to its vertebral articulation.

On Nov. 16 the involved rib was removed for a distance of about 3 in. (7 cm.) from its vertebral articulation.

The pathologist's report on the osseous tissue was as follows: The specimen (Fig. 4*B*) is a portion of rib measuring 7 by 1 by 1 cm. In the proximal portion of the rib there is an expanded area devoid of cortex and replaced by a grey hemorrhagic fungating mass, which on cross section is spongy and irregular. This tissue extends across the entire cortex. Microscopy shows the tumor tissue to be composed of an osteogenic neoplastic tissue which has destroyed the cortex and overlying periosteum. The new bone trabeculae are numerous and constitute about half of the mass. The cellular elements between trabeculae are dense and highly proliferative and show marked nuclear hyperchromatin, indicative of regenerative activity.

The tumor was diagnosed as an osteoid osteoma.

Osteofibroma.—Two cases of osteofibroma have been reported in the literature. They do not present any characteristic clinical or roentgenographic manifestations. The diagnosis of an osteofibroma is purely one of histopathology.

Fibrous Dysplasia.—Of the 205 collected cases, there were 35 cases of fibrous dysplasia of the rib, constituting 17.0% of all the tumors and 33.0% of the benign tumors. It occurred 32 times in men and 3 times in women. The ages of the patients ranged from 19 to 45 years, with a preponderance of patients between 20 and 30 years of age. (These statistics should not be taken too literally because 29 of the cases reported by Schlumberger were taken from the records of the Army Institute of Pathology, where most of the material reviewed is obtained from younger men.) In 11 of the 35 cases the fibrous dysplasia was asymptomatic and was an incidental finding on x-ray examination of the chest; in 20 cases the patients complained of

local pain of from one month to several years' duration, and in the other four cases no reference was made to symptomatology. Roentgenograms of the involved area do not reveal any specific changes. However, most commonly they show an expanded thinned-out area in the cortex of the rib, with the entire involved area radiotranslucent and irregularly traversed by denser bony trabeculae.

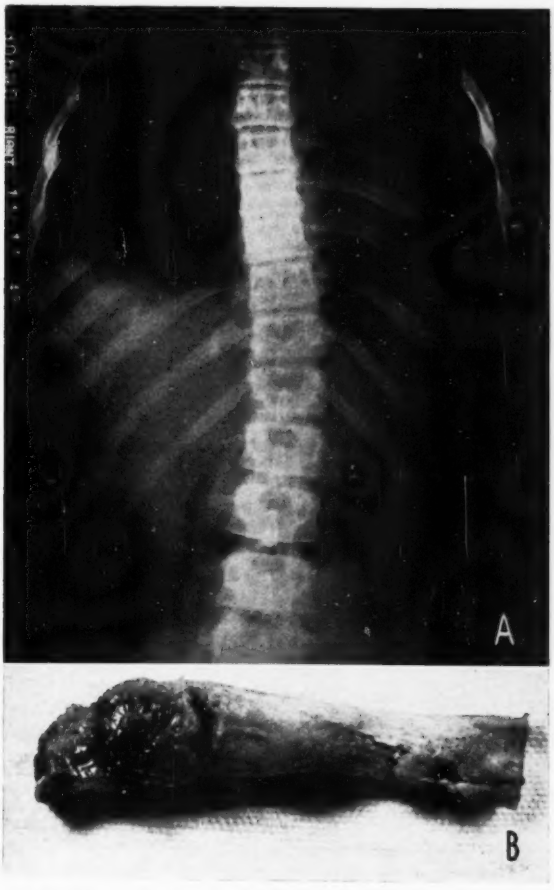

Fig. 4.—*A*, roentgenogram of the lower thoracic region, revealing a lumbodorsal scoliosis and an area of radiolucency in the medial portion of the 11th rib on the right side.

B, in the proximal portion of the rib there is an expanded dark friable area. On cross section the rib in that area was spongy and irregular—same case as *A*.

Grossly, these tumors are symmetrical fusiform expansions of the rib, with a thinned cortex. On cross section the inner and medullary area appears replaced by a yellowish-grey and pinkish rubbery substance. In some areas, especially toward the periphery of the abnormal tissue, firmer gritty material is encountered. The microscopy shows bundles and whorls of vascularized fibrous connective tissue and islands of cartilaginous and myxomatous tissue. Toward the periphery of the tumor bony trabeculae may be found extending into the fibrous connective tissue.

A 37-year-old man entered the hospital on April 2, 1944, complaining of pain on the right side of the chest, in the posterior axillary area, and in the upper part of the right side of the abdomen for three days. The pain had become progressively worse since its onset. The patient gave a history of having sustained an injury to the lower part of the right side of the chest about a month previously. The physical examination revealed a local area of swelling and tenderness in the anterior axillary area of the right side of the chest. Roentgenographic examination of the involved area (Fig. 5*A*) showed an expanded area along the lateral aspect of the seventh rib. The cortex of the rib was of increased density, while the medullary area was radiolucent.

On April 12, the fused sixth, seventh, and eighth ribs were removed as one tumor mass.

The pathologist's report on the excised tissue was as follows: The specimen (Fig. 5*B*) consists of a hard mass fixing three ribs into one neoplasm. The ribs and tumor measured 16 by 8 by 1.5 cm., in the center of which there is a 5 cm. elastic prominence over the ribs. Transversely, the mass is irregularly firm and soft and measures 4 cm. in diameter. Histopathologic studies show the tumor to consist chiefly of bundles and sheaths of very long spindle cells with

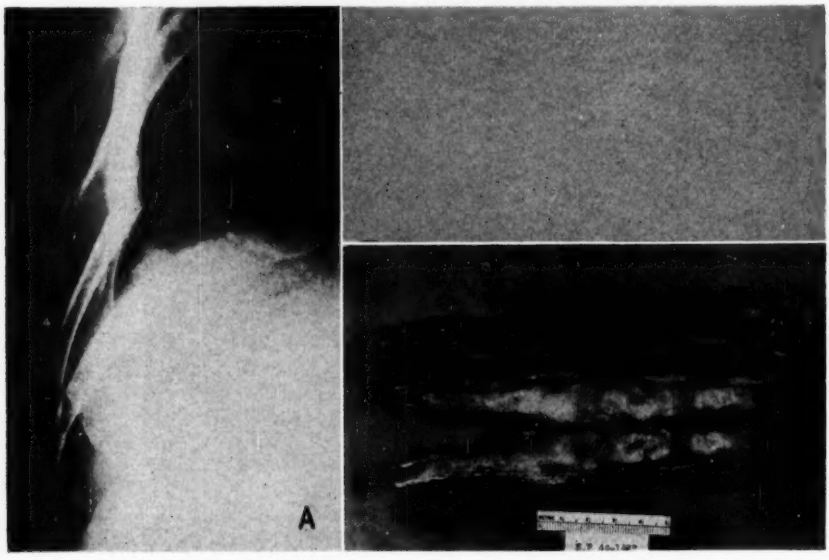

Fig. 5.—*A*, there is an expanded area along the lateral aspect of the seventh rib, the cortex of which is of increased density while the medullary portion of the rib is of lesser density. *B*, the tumor (S. P. 44-1462) seen in *A* consists of a hard mass fixing the sixth, seventh, and eighth ribs into one neoplasm. Seventh rib, shown in cross section, is comprised of a fusiform irregular greyish mass of elastic tissue.

wavy processes. These are arranged in an irregular manner, and scattered throughout are numerous newly formed blood vessels. The cells are equal in size; their nuclei are small, uniform, oval, and vesicular. Scattered throughout are the remains of bone trabeculae in various stages of solution. In their neighborhood are a number of osteoclasts. In other places there are fragments of cartilage together with some new bone formation.

A diagnosis of fibrous dysplasia was made.

Hemangioma.—There were eight vascular tumors of the rib encountered in this survey. These constituted 3.9% of all primary rib tumors and 7.6% of the benign tumors. The ages of the patients ranged between 21 and 61 years. The chief symptoms, as in other rib tumors, were pain and a mass. In two instances there was an accompanying cough, and in another there was some respiratory distress—in neither of these cases were the latter symptoms related to the neoplasm. The duration of

symptoms varied from 9 months to 15 years; in four cases they were of 2 to 3 years' duration before surgery was performed. The most constant physical finding was local tenderness over the site of the tumor. In these cases, x-ray examination of the involved area showed either of two patterns: (1) a nest of small radiolucent areas, separated by dense osseous tissue in an expanded process within the rib, having the appearance of a group of soap bubbles, or (2) a larger cystic area involving the greater part of an expanded area in a rib within which there were fine trabeculae of osseous density. These tumors have been described in all parts of the rib, but oftener they are located in the posterior part of the rib.

Grossly, these tumors are expanded areas in the rib with little or no erosion of its surface. The walls of the tumor are of variable density. On the cut surface one notes vascular networks of multilocular dilated blood sinuses separated from one another by bone and loose connective tissue. Histologically, these tumors are comprised of dilated vascular areas which contain blood and are lined by endothelium.

A 31-year-old woman entered the hospital on Aug. 28, 1951, complaining of pain in the upper part of the left chest of about two years' duration. Examination revealed a local tender zone in the upper paravertebral area of the left hemithorax. Roentgenographic examination of the chest disclosed a multicystic area in the broadened medial part of the third rib posteriorly (Fig. 6*A*) in proximity to the costovertebral area. The areas between the cysts were dense and osseous in nature.

On August 30 the posterior part of the third rib and tumor were removed.

The pathologist's report on the specimen was as follows: The tissue consists of a portion of rib measuring 6 by 2.5 cm. in greater diameters. It is hemorrhagic and gray-tan in color. On section it is hemorrhagic and irregularly cystic, soft and firm (Fig. 6*B*). Microscopy shows uniformly vascular channels of moderate size embedded in bony tissue. Each channel is lined by a single layer of flattened endothelial cells. The lumina of the spaces are filled with erythrocytes. Surrounding the channels there is a loose fibrous connective tissue. Bone spicules are distributed throughout the tissue in an irregular manner. These spicules show well-developed cancellous structure and some irregularity of the trabecular structure.

The diagnosis was hemangioma of rib.

Xanthomatous Giant-Cell Tumor.—In the collected series of cases 10 of the tumors were classified as giant-cell tumors. These constituted 4.9% of the total number of cases and 9.4% of the benign tumors. The ages of the patients with this condition varied from 9 to 49 years, with half of the patients being less than 17 years of age. As in other primary rib neoplasms, the chief symptoms were pain and tumor. In one instance there was an accompanying loss of weight; in another there were weakness, malaise, fever, and ulceration of the skin overlying the tumor. In seven of the cases the symptoms were less than one year in duration. Roentgenographic examination of the involved area usually shows a fusiform enlargement of the rib, with more or less sharp demarcation of the lesion from the neighboring osseous tissue. The periosteum over the tumor is fairly well preserved, although in some instances erosion of the cortex has been noted.

Grossly, these tumors may be comprised of one or more expanded areas. The surface of the tumor is of variable thickness. The color varies from a pinkish-red interspersed with yellow necrotic areas to a grey. On the cut section it appears rather vascular and contains prominent small cystic areas enclosing dark blood. The periphery of the tumor shows thinning of the bone. Central necrosis is not infrequent. On microscopic examination there is observed a more or less uniform distribution of spindle and multinucleated giant cells. The vascularity of the tumor is variable. Not infrequently one may observe double refractory lipid bodies.

Miscellaneous.—In this category of cases I have placed six cases which are rather uncommon—a fibroma of the rib reported by Sommer and Major, a myeloplax reported by Marottoli, a solitary myeloma reported by Aufses, a xanthoma contributed by Prevost, a myxoma removed by Fesani, and a chondromyxoid fibroma reported by Goorwitch. These cases are described in more detail in Table 1.

MALIGNANT TUMORS

Chondrosarcoma.—The malignant counterpart of the chondroma of the rib, chondrosarcoma, occurred in 36 cases, comprising 17.5% of all the rib tumors and 36.3% of the malignant tumors. The ages of the patients ranged from 16 to 74 years, with approximately two-thirds of the patients being between 32 and 60. As in the other primary rib tumors, the chief symptoms were pain and tumor on the chest wall.

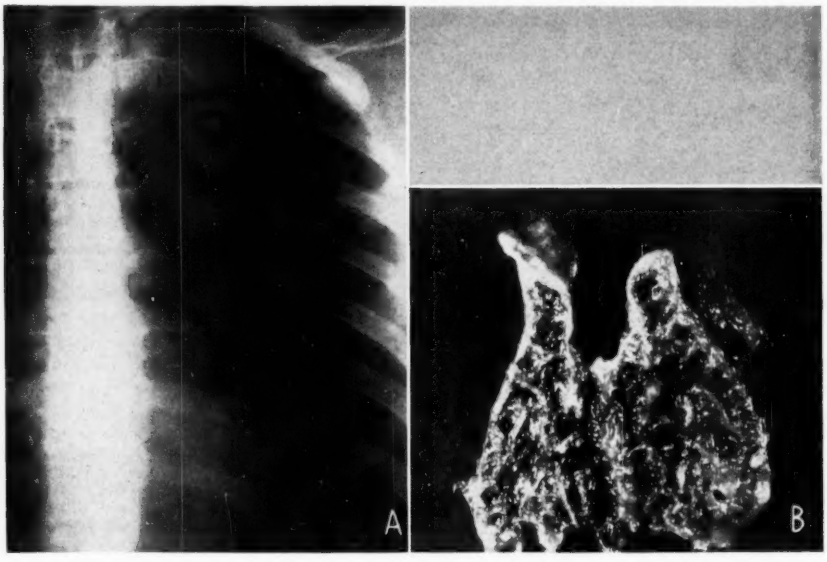

Fig. 6.—*A*, there is a soap-bubble-like expansion of the third left posterior rib. Between the cystic areas there is dense bony tissue.

B, cross section of the tumor (S. P. 51-1393) removed from the patient shown in *A*. Tumor consists of hemorrhagic and grey irregular cystic areas separated by soft and firm tissue.

The duration of symptoms before the patient came to the attention of the surgeon was 3 days to 15 years. Dyspnea was encountered in only four patients. In the majority of cases a tumor could be palpated when the patient was seen for the first time. Roentgenographic examination of the affected area usually shows an opaque mass fixed to the ribs. Within the substance of the tumor there may be noted areas of greater (bony) density and/or areas of radiolucency due to degenerative changes in the tumor. In approximately half the cases the tumor was located in the anterior chest wall in or near the costal cartilage.

Grossly, these tumors vary in size from about 5 cm. in diameter to that described by Condon and Harper which was 30 cm. in diameter. The surface of these tumors is bluish-grey in color, nodular, and often surrounded by a pseudomembrane. Areas of necrosis and softening of the nodules are not uncommon. The cut surface of the

tumor is often the same color as its exterior. However, cyst-like areas of degeneration filled with mucoid material are not uncommon. In some fields the microscopy is not unlike that of the benign chondroma. However, by studying the entire tissue one notes areas of less uniformity and greater irregularity in cellular arrangement. In addition, the cells show nuclear atypism, with many cells and nuclei varying in size, shape, number of nuclei, and degree of nuclear activity.

A 32-year-old man entered the hospital on Feb. 15, 1945, with a history that four years previously a small painful mass developed on one of his ribs in the lower part of the right side of the chest posteriorly. At that time a piece of the tumor was removed (in Chile) and reported as "chondroma." Sometime thereafter the tumor began to increase in size and to give rise to persistent pain locally. Removal of the tumor a year prior to the present admission, by the same surgeon, showed the neoplasm to be a "benign chondroma." Four months later signs of involvement of the hip developed on the same side. At the time of the present admission to the hospital he had paralysis of the lower extremities bilaterally and a large mass fixed to the lower part of the chest and vertebral column. X-rays of the chest, spine, and hip showed extensive destructive changes in these areas. No attempt was made to remove the tumor completely. However, a piece of tissue removed for biopsy showed the presence of a chondrosarcoma. The patient died some months later, four years after the onset of illness.

Osteogenic Sarcoma.—These tumors (eight cases) comprised 2.9% of all the primary rib tumors and 6.0% of the malignant tumors. The ages of the patients, as given in five cases, were 27 (in two cases) 32, 44, and 54 years, respectively. As in the other cases of primary rib tumors, the initial symptoms were pain and tumor. In three cases dyspnea was also present; in one, palpitation; in another, deformity of the chest wall, and in another epigastric pain. The duration of the symptom-complex in the five cases in which it was recorded was one week (two cases), nine months, one year, and two years, respectively. Roentgenographic examination of the involved area showed a neoplasm not unlike that of a chondrosarcoma in size, location, and configuration. However, it was denser and contained more sclerotic areas than the chondrosarcoma.

Grossly, these tumors are not as bulky as the chondrosarcoma and may involve one or more ribs. The surface of the tumor is reddish-grey, hard, and irregular. The periosteum is rough and contains bony spicules. Necrosis and destructive changes in the rib and tumor are not uncommon. When the tumor arises from the medullary portion of the rib, it is softer than when its origin is from the periosteum. Histologically, these tumors are composed of compact anaplastic bony tissue with pleomorphic spindle, polyhedral, and giant cells. The cells are often multinucleated.

Fibrosarcoma.—There were 12 cases of fibrosarcoma, comprising 5.8% of all the tumors and 12.1% of the malignant tumors. The ages of the patients ranged from 10 to 61 years, with about 60% of the patients being between 34 and 50 years. The symptom-complex was not unlike that encountered in patients with other primary rib tumors. In one patient, reported on by Blades and Paul, the symptoms were malaise, elevation in temperature, and pain. The duration of symptoms in the 12 cases varied from three weeks to four and a half years. The x-ray manifestations of a fibrosarcoma of the rib are not unlike those of a chondrosarcoma (in size, shape, location, and rib involvement) except, perhaps, that in fibrosarcoma the anterior part of the rib is not as commonly involved as in chondrosarcoma.

Grossly, they are fusiform tumors, involve one or more ribs, and have a rubbery hardness. Whereas the outer surface may be hemorrhagic, the cut surface is usually pale grey in color. Microscopy reveals variegated attenuated cell formations of immature spindle cells containing relatively large nuclei.

A 36-year-old woman entered the hospital on Aug. 16, 1949, complaining of pain in the left axillary area and a mass in that region (about 18 months previously she had a tumor removed from that area in another hospital). At the time of the present hospital admission there was a healed scar in the left axillary area deep to which there was an irregular firm tender mass about 4 in. (10 cm.) in length which was fixed to the ribs. Roentgenographic examination of the chest showed an irregular opacity along the anterolateral part of the left hemithorax, disruption of the continuity of the left fifth rib, and previous surgical removal of the sixth and seventh ribs in the same area (Fig. 7*A*).

On Aug. 22 the neoplasm with the surrounding tissues, including the fifth to the eighth ribs, and the accompanying intercostal muscles were excised.

The pathologist's report on the specimen was as follows: The tissue consists of portions of four ribs measuring up to 16 cm. in length (Fig. 7*B*). On the convex surface there is a firm mass measuring 13 by 5 by 4 cm. The external surface is dark brown and covered by a fibrous capsule. The concave surface has two soft globular masses which are adherent to one another and appear to arise from the ribs. On cut surface each mass contains serohemorrhagic fluid in cystic inclusions. The walls of the cystic cavities are covered by lobulated soft yellowish-grey

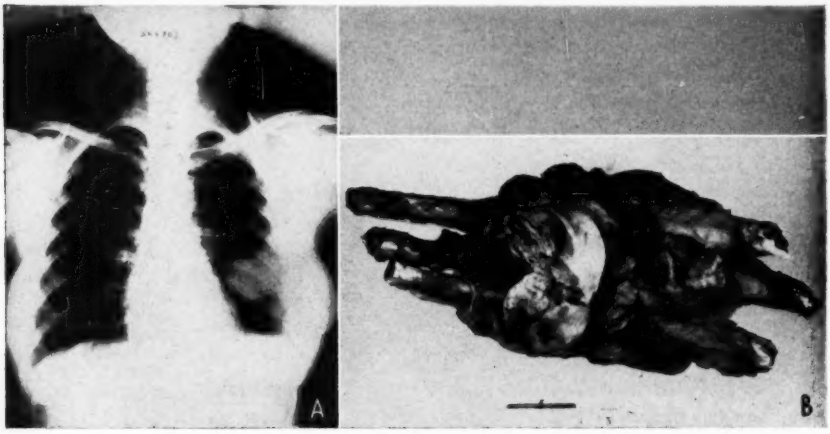

Fig. 7.—*A*, there is a large opacity in the anterolateral portion of the left hemithorax, extending from the fourth to the sixth ribs in that area. *B*, tissue (S. P. 49-3987) removed from the patient whose roentgenogram is shown in *A* consists of portions of the fifth through the eighth ribs with the interposed intercostal muscles. On the concave surface of the mass there are two soft globular grey masses, which on the cut section reveal the presence of serohemorrhagic fluid in a fibrotic casing.

tissue, similar to that of the tumor itself. Microscopy of representative tissue shows the tumor to consist of spindle-shaped and irregularly polygonal cells in varying sizes. In some of these cells the nuclei are round and oval and vascular; in others they are irregular and hyperchromatic. Mitotic figures are fairly numerous. The tumor cells are arranged in interlacing bands. In some places the tissue is loose, edematous, and fragmented, while in others it has an angiomatous element with engorged vessels and foci of hemorrhage.

The diagnosis was fibrosarcoma.

There was recurrence three times, and on each occasion a mass was removed. At the time of the third recurrence the mass involved the vertebral column, and only small fragments of this tumor could be removed. The patient is alive, but not well, five years after the onset of initial symptoms.

Mixed Sarcomas.—There were eight cases of mixed cell sarcomas of the rib. These were (1) osteogenic myxofibrosarcoma, (2) chondromyxosarcoma, (3) osteogenic myxochondrosarcoma, (4) chondroosteosarcoma, (5) osteochondromyxo-

sarcoma, (6) osteochondrosarcoma, (7) fibrochondrosarcoma, and (8) fibromyxomatous osteosarcoma. Little can be said about these cases, for they present nothing which is distinctive.

Endothelioma (Ewing's Tumor).—This tumor was encountered in 24 cases—in 11.7% of the entire series of primary rib tumors and in 24.2% of the malignant tumors. The youngest subject was 5 years of age and the oldest 31 years, with 67% of the patients being between 10 and 20 years of age. Notoriously, the symptoms were of short duration and relatively rapidly progressive. The shortest duration of illness was 15 days, while the longest period was 18 months, being 6 months or less in 17 cases and 3 months or less in 10 cases. Here, too, the chief symptoms were pain and/or neoplasm. Physical examination most frequently revealed a tender area or mass fixed to the chest wall which did not differ from that of any other tumor of costal origin. Roentgenographic characteristics of this condition are variable. In some instances there is a diffuse expanded bone lesion with little periosteal reaction and irregular absorption of the medullary area, as is often seen in a subacute osteomyelitis. In other instances there is widening of the medulla and increased density of the widened cortex, and the periosteum appears veiled. If periosteal reaction takes place, it often assumes a laminated or "onion peel" appearance. While the tumor process is usually confined to one rib, it is not uncommon for more than one rib to be involved in the growth.

Grossly, these tumors are bulky, lying in the subperiosteal tissues, and are covered by a periosteal membrane. On cross section the medullary cavity is narrowed and appears encroached upon by the widened subperiosteal and endosteal laminated bone. The histology of this tumor is fairly constant: The cells are often small and compact; round, oval, or polyhedral; in the form of sheets, and with round or oval nuclei. The cytoplasm is often scant; mitosis is not infrequent; osteoclasts are occasionally present, and the vascularity is variable.

A young man, aged 20 years, entered the hospital on March 15, 1949, complaining of pain in the lower part of the back and in the right thigh of two months' duration. On awakening one morning he suddenly had a dull ache in the upper lumbar area. A few days later he noted that the pain extended downward to the iliac crest and to the thigh on the right side. The physical examination was essentially noncontributory except for a tender mass on the posterior aspect of the right lower chest wall which appeared fixed to the 11th rib. X-ray studies of the local area revealed thickening and irregularity of the posterior part of the 11th rib (Fig. 8*A*).

On March 18, 1949, the tumor, with the 11th and the involved 10th rib and the surrounding soft tisues, was removed.

The pathologist described the tissues as follows: The specimen (Fig. 8*B*) consists of portions of two ribs, each measuring 12 by 1.5 cm., with an attached globular mass measuring 8 by 5 by 4 cm. The external surface is covered by a thin capsule. It is red-brown and pink-tan in areas. On the cut surface this mass is fleshy in consistency, tan and grey in color, and irregularly lobulated. Some areas are hemorrhagic, soft, and necrotic. The tumor mass involves both ribs but chiefly the 11th rib. Microscopy revealed the tumor to consist of round, polygonal, and ovoid cells, varying but slightly in size. The nuclei are round, large, and in places vesicular. Many nuclei are hyperchomatic, and some are in mitotic division. The cells are often arranged in sheets or nests and in places in alveolar formation. The stroma is scanty and vascular.

The diagnosis was endothelioma of the rib.

Malignant Giant-Cell Tumor.—Unlike similar tumors of other osseous tissues, there is but one case of a malignant giant-cell tumor of a rib reported in the literature, the one reported by Guglielmini and Pironti.

Miscellaneous Malignant Tumors.—There were 12 cases in this category, which may be subdivided into four groups. In the first group there were seven cases: three round-cell sarcomas, two lymphosarcomas, one small round-cell sarcoma, and one lymphocytoma. It seems to me that all of these tumors are lymphosarcomas, with only a minor difference in the description of the cell comprising the tumor. The second group consists of two plasmocytomas. These are a distinct entity of neoplastic cells. They are similar to plasma cells and may be placed in the category of the solitary myeloma tumor of bone-marrow origin. The third group similarly consists of two cases, of reticulum-cell tumors, and these are also a recognized entity of reticulum-cell sarcoma. The fourth group consists of a single case of hemangioendothelioblastoma, reported by me.

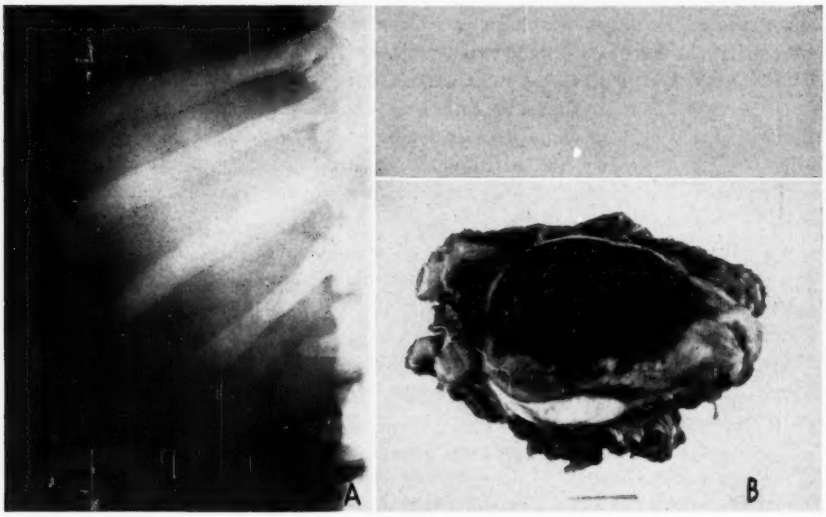

Fig. 8.—*A*, there is a localized area of periosteal thickening and irregularity along the posterior part of the 11th rib.

B, tumor (S. P. 49-1763) removed from the patient whose roentgenogram is shown in *A* consists of the 10th and 11th ribs, with a beefy irregular globular mass, fixed to these ribs. On cross section the tumor contained hemorrhagic, necrotic, and soft gray tissue.

As in all other cases of primary tumors of the rib, tumor and pain were the two outstanding symptoms in these cases, too. The ages of the patients varied from 9 to 63 years. The duration of symptoms varied from three weeks to six years, with 60% of the patients in the known cases being symptomatic for six months or less before coming to the attention of the physician.

A 49-year-old man entered the hospital on Nov. 8, 1943, complaining of a swelling on the left side of the chest of three weeks' duration. Physical examination revealed a mass overlying the region of the seventh rib in the posterolateral part of the left hemithorax. It was firm, nontender, and fixed to the underlying ribs. An x-ray of the chest (Fig. 9*A*) showed an irregular opacity along the upper posterolateral chest wall, with indentation of the lung bed at the level of the fifth, sixth, and seventh ribs. Oblique x-ray studies showed destruction of the sixth rib in the axillary area.

On Nov. 17, the involved fifth, sixth, and seventh ribs with the surrounding soft and osseous tissues were removed en masse.

The pathologist's report of the tissue removed was as follows: The mass (Fig. 9*B*) sent to the laboratory consists of an irregularly firm and red tissue in which there are three ribs embedded. The tumor mass is 13 cm. in length and 7 cm. in width. The intervening intercostal muscles between the resected ribs are involved in the neoplastic process. On the cut section the tissue is red and pale grey with local hemorrhagic and necrotic areas. Microscopy discloses a variegated picture. In one preparation there are broad bands of dense fibrous tissue through the growth, with islands of infiltration of vascular elements. The latter consists of plump spindle-shaped and irregularly polygonal cells of varying size. The nuclei in many cells are vesicular and in other areas quite irregular and hyperchromatic. Occasional blood channels lined by immature endothelial cells are noted, and in other areas the channels are lined by cells similar to the tumor cells.

The diagnosis was hemangioendothelioblastoma.

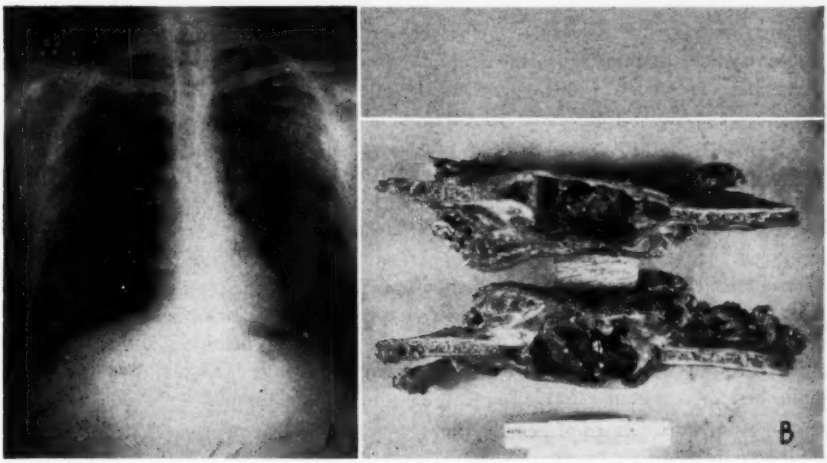

Fig. 9.—*A*, there is an irregular opacity in the region of the sixth rib posteriorly on the left side. There is partial destruction of the rib in that area.

B, tumor mass (S. P. 43-3844), removed from the patient whose roentgenogram is seen in *A*, consisting of three ribs in the central portion of which there is a necrotic mass. Between the ribs the infiltrated intercostal muscles can be seen.

COMMENT

Primary tumors of the rib are not quite as uncommon as one would gather from the relatively few cases reported in the literature. Obviously, the paucity of cases is due to failure to report small series of cases.

The most frequent symptoms encountered in patients with primary rib tumors, regardless of whether the growth is benign or malignant, are pain and swelling, either as individual symptoms or together. Therefore one cannot draw any conclusions as to the nature of the tumor simply from symptoms. The age of the patient is no criterion upon which to base a diagnosis of benign or malignant growth, although endothelioma, round-cell sarcoma, plasma-cell sarcoma, and lymphosarcoma are distinctly commoner in patients in the younger age group, i. e., those below 30 years of age. The tendency is for the patient with a malignant tumor to seek medical care earlier than one who has a benign tumor because of the more rapid expansion of the tumor and more persistent and/or severe pain in the local area of the growth. There-

fore any patient with an enlarging tumor of the rib and persistent and/or severe pain in the local area should be suspected of having a malignant tumor of the rib until proved otherwise.

The roentgenograms of tumors of the ribs are not conclusively characteristic of their pathology. However, in a general way it may be said that a tumor confined to a single rib is more likely to be benign than malignant. At the same time it must be remembered that at its inception a malignant tumor, too, is confined to a single rib, and therefore a roentgenogram of the rib at this stage will show but a single rib involved. Thus, the presence of a tumor in one rib does not necessarily imply a benign tumor. That benign tumors of the rib may involve more than a single rib is well illustrated in the case reported by Poppe and Berg. Consequently, a definitive pathologic diagnosis cannot be made from x-ray studies of the ribs alone.

In reviewing the literature on the subject, one gains the impression that many writers believe that most primary rib tumors are malignant, a view to which I do not subscribe. However, I do believe, as do others, that many of these tumors are potentially malignant and should be treated as such. The removal of a piece of the rib for biopsy purposes has little or no justification. If the patient's condition is good enough to permit exploration for the removal of a piece of rib, it is generally good enough for more radical excision. If the patient's condition permits, all the tissues involved in the neoplastic process and in proximity to this process should be removed, including the bone, cartilage, periosteum, muscle, nerves, vessels, and even pleura and lung if necessary. It is better to follow the principle that when in doubt the tissue should be removed than to leave any suspicious tissue behind. At the time of surgical intervention one may encounter a well-delimited and even encapsulated tumor which has all the gross characteristics of a benign tumor but which upon histologic examination is found to be a malignant tumor.

In a number of cases of primary tumors of the rib there is reason to believe that a preexisting benign tumor antedated the malignant lesion. Condon and Harper report a case in which a tumor of the rib had been present for 13 years before it was removed and proved to be malignant. Janes reports a similar case with a 15-year history. Lentino reports a case in which 20 years passed before the tumor was shown to be malignant. In this case, three years after onset of symptoms, a chondroma of the rib was removed. After a number of recurrences and removal the tumor was diagnosed as an osteochondrosarcoma. Just as one is often unable to determine the nature of the tumor by simple gross inspection, so too may one be unable to determine the degree of maturity by simple microscopic examination of a single section. After removing a tumor from a rib, R. E. Johnson submitted sections of the tumor to a number of pathologists for review. The reports of these pathologists are interesting: Fifty-four called the tumor an osteogenic sarcoma, 6 called it a chondrosarcoma, 27 believed it to be an osteoid osteoma, 3 thought it was a fibrous dysplasia, 4 diagnosed it as healing bone with foreign-body reaction, 1 described it as osteoid tissue forming a tumor, and another thought it was an aneurysmal bone cyst. If these pathologists were at such variance in their diagnosis, it is readily understood why the surgeon is frequently unable to make a definitive diagnosis at the operating table of benign or malignant growth. It may also account for the number of instances in which "benign" tumors have been described as having undergone "malignant" transformation soon after the original extirpation.

CONCLUSIONS

1. Primary rib tumors, as reported in the literature, are about as frequently benign as malignant.

2. The likelihood of malignant transformation of a benign rib tumor is frequent enough to justify wide removal as soon as discovered.

3. Whether a tumor is benign or malignant is often a difficult decision unless careful and detailed studies are made grossly and microscopically.

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LEIOMYOMA OF THE ESOPHAGUS

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A WIDE variety of benign neoplasms may arise from the esophagus, including leiomyomas, fibromas, hemangiomas, adenomas, polyps, papillomas, mucoceles, neurofibromas, myxofibromas, and lipomas. It is well recognized that these lesions are rare. A careful review of the literature serves to confirm this impression.[1]

In examining the literature from 1717 to 1932, Patterson [1a] found only 62 cases of benign tumors of the esophagus, all incidental postmortem findings. Adams and Hoover [1c] added 26 additional cases from 1933 to 1943, bringing the total to 91 cases.

There has been a relative increase in the number of cases recorded since 1944, with Harrington and Moersch [1p] reporting 15 benign esophageal tumors among 11,000 patients complaining of dysphagia. These same authors found 44 benign tumors of the esophagus, of which 32 were leiomyomas, in a series of 7,459 postmortem examinations. Schafer and Kittle,[1f] in 1947, surveying 6,001 autopsies at the University of Chicago, The School of Medicine, noted 11 benign neoplasms of the esophagus.

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Chi and Adams,[1k] in 1950, reported two benign tumors of the esophagus, in contrast with 246 malignant tumors found clinically from July, 1936 to July, 1947. These authors in reviewing the literature from 1912 to 1948 found 209 benign tumors of the esophagus, of which 46 were leiomyomas.

Obvious discrepancies and omissions are present in the literature regarding type, number, and incidence of benign neoplasms of the esophagus. One must take into consideration the small number of clinics which have reported their postmortem tumors of the esophagus. This is emphasized in Table 1, where a total of 67 leiomyomas of the esophagus are reported, of which 43 were found at necropsy.

TABLE 1.—*Leiomyoma of the Esophagus Reported to Date*

Author	Date	No.	Post Mortem
Patterson	1912-1936	6	6
Ohsawa	1933	1	..
Harrington and Moersch	1944	32	32
Harrington and Moersch	1944	2	..
Harper and Tiscenco	1945	1	..
Calmenson and Clagett	1946	1	..
Sweet	1947	1	1
Schafer and Kittle	1947	1	..
Kenworthy and Welch	1948	1	..
Harrington	1948	4	..
Hurwitz	1949	1	..
Nissen	1949	1	..
Daniel and Williams	1950	1	..
Dugan and Meagher	1950	1	..
Chi and Adams	1950	1	..
Bigger, Kay, and Vinson	1950	1	..
Cornell	1950	5	4
Madden and Olmstead	1950	1	..
Goldman and Masters	1950	2	..
Garlick and Stegmaier	1951	1	..
Myers and Bradshaw	1951	1	..
Moore and Wilder	1952	1	..
Total		67	43

The surgical attack on benign tumors of the esophagus has shown a dramatic increase in the past decade. This is well illustrated upon reviewing cases of leiomyoma of the esophagus.

In 1947 Schafer and Kittle [1f] found only three patients with esophageal leiomyoma surgically treated, one of whom recovered. In 1950 Goldman and Masters [1j] mentioned eight patients with this condition, six of whom recovered. Bradshaw and Myers,[1o] in 1951, reported on 16 patients (with surgical enucleations only), with 1 death.

Table 2 shows only 1 patient approached surgically prior to 1944 and 23 patients treated surgically since 1944.

REPORT OF A CASE

In the following case a correct diagnosis was made prior to operation, and this case is added to the growing list of surgically managed leiomyomas of the esophagus.

This 52-year-old white man was in good health until 10 months prior to hospitalization, at which time he noted the onset of a burning sensation in his midepigastrium which radiated to the right and upper left quadrant and was constant throughout the day and increased during

TABLE 2.—*Operative Cases of Leiomyoma of the Esophagus*

Author	Date	Age, Yr.	Sex	Symptoms	Location	Procedure	Result
Ohsawa	1933	43	F	Dysphagia	Lower	Enucleation	Satisfactory
Harrington and Moersch	1944	56	F	Substernal pain	Lower	Excision	Satisfactory
Harrington and Moersch	1944	39	M	None	Lower	Esophagogastric resection	Satisfactory
Harper, Tiscenco, and Calmenson	1945	41	M	Vomiting	Middle	Esophagogastric resection	Died
Clagett	1946	52	M	Dysphagia, substernal distress	Upper middle	Enucleation	Satisfactory
Schafer and Kittle	1947	46	M	Epigastric pain, nausea, vomiting	Lower	Resection of wall	Satisfactory
Kenworthy and Welch	1948	56	F	Anorexia, vomiting	Cardioesophageal junction	Esophagogastric resection	Satisfactory
Nissen	1949	56	M	Dysphagia	Middle	Enucleation	Satisfactory
Hurwitz	1949	31	M	None	Middle	Enucleation	Satisfactory
Harrington	1949	31	M	None	Middle	Resection of wall	Satisfactory
Harrington	1949	58	F	Dysphagia, regurgitation	Lower	Enucleation	Satisfactory
Harrington	1949	53	M	Cough, chest pain	Lower	Resection of wall	Satisfactory
Harrington	1949	19	F	Choking, dysphagia	Cardioesophageal junction	Esophagogastric resection	Satisfactory
Goldman and Masters	1950	63	F	Dysphagia	Lower	Enucleation	Satisfactory
Goldman and Masters	1950	51	F	Dysphagia	Lower	Esophagogastric resection	Satisfactory
Daniel and Williams	1950	88	F	None	Lower	Enucleation	Satisfactory
Bigger, Kay, and Vinson	1950	24	M	Dysphagia	Middle	Enucleation	Satisfactory
Chi and Adams	1950	25	M	Heartburn	Middle	Esophagogastric resection	Satisfactory
Dugan and Meagher	1950	27	F	Dysphagia, epigastric pain	Middle	Enucleation	Satisfactory
Madden and Olmstead	1950	28	M	Cough, chest pain	Upper	Enucleation	Satisfactory
Cornell	1950	54	M	Indigestion	Lower	Enucleation	Satisfactory
Garlick and Stegmaier	1951	48	M	Substernal pain	Middle	Enucleation	Satisfactory
Myers and Bradshaw	1951	40	M	Regurgitation	Lower	Enucleation	Satisfactory
Moore and Wilder	1952	52	M	Epigastric discomfort, weight loss	Middle	Enucleation	Satisfactory

Fig. 1.—Preoperative esophagram demonstrating typical x-ray appearance of leiomyoma in midthird of esophagus.

Fig. 2.—Postoperative esophagram demonstrating normal-appearing esophagus.

the night. This condition was unrelieved by food, change in activity, or change in position. The patient experienced no nausea, vomiting, malaise, constipation, melena, red blood in stools, acholic stools, or change in color of urine. There were no chills, dysuria, urgency, or frequency. There was no cough, chest pain, or ankle edema. There was a 7 lb. (3.2 kg.) weight loss.

Past History.—A subtotal thyroidectomy was done in 1945 for chronic thyroiditis. A systemic review was noncontributory.

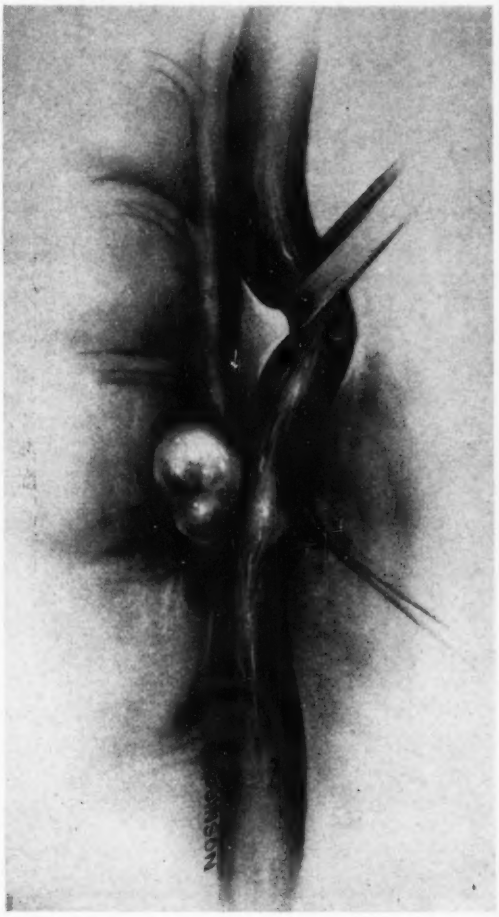

Fig. 3.—Artist's drawing showing typical appearance of tumor at time of surgery.

Physical Examination.—The patient's temperature was 37.3 C., pulse rate 90, respiration rate 20, and blood pressure 140/80. The positive physical findings were as follows: The patient was a tall, wiry, asthenic type, showing evidence of recent weight loss. In the neck region there was a well-healed collar incision. Examination of the chest revealed nothing abnormal. The liver, spleen, and kidneys were not palpable. No tenderness or abnormal masses were noted except a 2 by 1 cm. superficial freely movable mass just above the umbilicus in the midline, suggestive of a lipoma.

Laboratory Findings.—The urine was acid, with a specific gravity of 1.023. Blood studies showed 4,800,000 red cells, 12.5 gm. of hemoglobin per 100 cc., and 7,200 white cells, with a differential distribution of 76% leucocytes, 18% lymphocytes, 5% monocytes, and 1% basophiles. The value for total protein was 6.8 gm. per 100 cc., with a normal albumin : globulin ratio. The blood urea nitrogen was 14 mg., calcium, 10 mg.; phosphorus, 2.8 mg., amylase 6.2 units, and cholesterol, 326 mg. per 100 cc. The guaiac reaction of stools was 1+ four times and 0 twice.

Fig. 4.—Photomicrograph demonstrating typical benign smooth muscle cells characteristic of leiomyoma.

The prothrombin time was normal. Results of gastric analysis were normal. The vital capacity was 4 liters on two determinations. The electrocardiogram was normal. Serial x-rays of the gallbladder and barium enema (with air-contrast study) showed nothing abnormal. A gastrointestinal series and barium swallow (Fig. 1) revealed a defect of the middle esophagus, suggestive either of extrinsic pressure or of an intramural lesion. A deformed duodenal bulb compatible with old ulceration was also present. There was no evidence of ulceration of the lesser curvature of the stomach. Bronchoscopy revealed no intrinsic lesion. On esophagoscopy at

30 cm. from the upper incisors, a bulging mass presented itself arising from the right lateral wall. The mucosa was smooth over this without ulceration or redness. Aspirated material stained by the Papanicolaou technique contained no malignant cells (1+). No biopsy specimen was taken. The impression was leiomyoma of the midesophagus. Chest x-rays showed nothing significant.

Operation.—The chest was entered by removing the right fifth rib, and a freely movable mass 2.5 by 1.5 by 1.5 cm. was noted in the midportion of the esophagus, as anticipated from preoperative studies. The pleura was incised over this mass, exposing the esophagus, and with use of careful dissection, the esophagus mobilized. A small longitudinal rent was made overlying the mass and the muscle of the esophagus reflected. The tumor was seen to be white and was firm throughout in consistency. It was shelled out from beneath the esophageal muscle without difficulty (Fig. 3). During the process of the dissection, the mucosa which was lying beneath the mass was identified, and at no point was it entered or injured. Several interrupted nonabsorbable surgical (silk) sutures were taken through the muscle, obliterating the defect. A catheter was placed in the chest and closure was effected.

Postoperative Course.—The patient's postoperative course was uneventful. He was given penicillin, 400,000 units daily, and dihydrostreptomycin, 0.5 gm. twice daily. The administration of fluids was started on the second postoperative day. The patient was ready for discharge by the seventh postoperative day.

Pathology Report of Operative Specimen.—A globular tumor measuring 2.5 by 1.5 by 1.5 cm. in diameter was present. This was firm in consistency, slightly lobulated, and yellowish-gray in coloration. On multiple cross sections it was relatively homogeneous, firm to palpation, and whitish-grayish-yellowish in color. There appeared to be fine white septi running through it (Fig. 4). Microscopic section of the tumor revealed it to be smooth-muscle cells, elongated, and showing very little variation in size. There was no anaplasia. A Masson trichrome stain showed the cells in the tumor to take the color of smooth muscle rather than of connective tissue.

Diagnosis.—The diagnosis was benign leiomyoma of the esophagus.

Follow-Up Report.—When seen two months later, the patient was completely free of complaints and was leading a normal life.

COMMENT

Benign tumors of the esophagus may be divided into two main groups: mucosal and extramucosal. Leiomyomas arise from the smooth muscle of the esophagus and are classified as extramucosal and intramucosal. These tumors are asymptomatic until they produce enough extrinsic pressure to cause varying degrees of obstruction, characterized by regurgitation, dysphagia, gurgling, nausea, vomiting, weight loss, and weakness. Ulceration, secondary to the pressure, may in addition cause pain, substernal discomfort, and bleeding.

The differential diagnosis necessitates ruling out other lesions of the esophagus and pathology elsewhere in the thoracic or the abdominal cavities. The clinical picture found in carcinoma of the esophagus has been well reviewed in the literature. The final diagnosis of leiomyoma of the esophagus rests on roentgenological examination, esophagoscopy, and biopsy, or surgical removal.

The x-ray picture (Fig. 1) is characterized by a noninvasive, smooth, often half moon-shaped filling defect following barium swallow. The tumor may originate anywhere in the esophagus. Esophagoscopy serves mainly to rule out an intrinsic lesion and to confirm the presence of extrinsic pressure. A biopsy through the mucosa carries with it the risk of mediastinitis and emphysema. It is safer in such instances to forego biopsy and proceed with surgery.

Surgical consideration demands careful preoperative evaluation, knowledge of the pathology, and thorough postoperative care. The decision to approach the esophagus from the right or left chest will depend on the location of the lesion. A most important factor is awareness of the ease with which these neoplasms can be enucleated without entering the mucosa and without subjecting the patient to unnecessary resection of the esophagus!

SUMMARY

Reports of leiomyoma of the esophagus, a relatively rare lesion, are now appearing more frequently in the literature. Errors in diagnosis can be averted by adequate clinical evaluation, roentgenology, and esophagoscopy. Enucleation can be performed with ease in the majority of cases and is the procedure of choice.

A case report of the successful surgical removal of an esophageal leiomyoma is presented, together with a review of the literature.

Case Reports

SURGICAL REESTABLISHMENT OF FALLOPIAN TUBAL PATENCY BY MODIFIED SOVAK TUBOPLASTY

Use of Polyethylene Tubing and Tubal Implantation

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AND
ABRAHAM TAMIS, M.D.
NEW YORK

RECONSTRUCTIVE pelvic surgery utilizing polyethylene (plastic) tubing is a new method offering increased possibilities for the cure of sterility due to obstructed, partially resected, or even absent Fallopian tubes. Closed Fallopian tubes constitute one of the major causes of female sterility. The prognosis in such cases need no longer be completely hopeless, even though repeated tubal insufflations, pelvic diathermy, or other conservative measures of therapy have failed. Increasing numbers of cases of successful reestablishment of tubal patency following reparative surgery are now being reported.[1] The hazards from pelvic or tubal reparative surgery have been minimized through present-day understanding of reproductive physiology, chemotherapy, antibiotics, and improved surgical techniques. Results of such surgery have also improved. Therefore, tubal surgery should be resorted to if the risk and limitation of the operation have been clarified to the patient and found acceptable.

The best results with reconstruction of Fallopian tubes have been obtained in tubes with occluded fimbriated ends. Complete occlusion of segments of the tube offer a much more difficult problem, which is best solved by the type of operation devised by Sovak.[1h] Patients who might benefit from such surgery should be informed of the possibilities.

In 1950 one of us (M. P. W.) was stimulated to experiment with the use of polyethylene tubing after a verbal communication with one of the pioneers in this

Read at the Clinical Society Meeting at the Jewish Memorial Hosiptal, New York, Feb. 5, 1952.

1. (*a*) Novak, E.: Congenital Anomalies of the Female Generative Organs, in Text Book of Gynecology, Ed. 3, Baltimore, Williams & Wilkins Company, 1948, pp. 121-125. (*b*) Westman, A.: Etiology, Diagnosis and Surgical Treatment of Female Sterility, in Greenhill, J. P., Editor: The 1951 Year Book of Obstetrics and Gynecology, Chicago, Year Book Publishers, Inc., 1951, p. 313. (*c*) Ahumada, J. C.; Chevalier, R. M., and Mezzadra, J. M. E.: Tubal Surgery and Sterility, ibid. pp. 314-315. (*d*) Milnor, G. C.; Bowles, H. E., and McCorriston, C. C.: Plastic Restoration of Fallopian Tubes Following Tubal Sterilization, in The 1950 Year Book of Obstetrics and Gynecology, Greenhill, J. P., Editor: Chicago, Year Book Publishers, Inc., 1950, pp. 356-357. (*e*) Rutherford, R. N.; Lamburn, H. M., and Banks, A. L.: Surgical Treatment of Sterility, Am. J. Obst. & Gynec. **58**:673-683 (Oct.) 1949. (*f*) Meigs, J. V.: Surgeon Surveys the Sterile Pelvis, Fertility & Sterility **1**:101-102 (March) 1950. (*g*) Beecham, C. T., and Sigman, M.D.: Technic for Re-Establishing Tubal Patency, in The 1949 Year Book of Obstetrics and Gynecology, Greenhill, J. P., Editor: Chicago, Year Book Publishers, Inc., 1950, pp. 377-378. (*h*) Holden, F. C., and Sovak, F. W.: Reconstruction of Oviducts: Improved Technic with Report of Cases, Am. J. Obst. & Gynec. **65**:150-157 (Jan.) 1953.

field, Dr. John Rock, who described a successful result. This paper presents our successful experience with a patient in whom temporary reestablishment of tubal patency was accomplished by tubouterine surgery, using polyethylene tubing as a temporary catheter. In our case additional features of interest which also contributed to the sterility were unusual anatomic findings of congenital pelvic developmental anomalies. It is not possible to diagnose or correct sterility due to anomalies except by exploratory laparotomy. Our operative findings and technical procedures will be fully discussed and illustrated.

REPORT OF A CASE

S. F., aged 30, white, Jewish, married four years, a nullipara, secundigravida, was first seen on Aug. 24, 1948, by M. P. W. with a complaint of secondary sterility. Her husband, aged 35, was found to have a highly adequate semen specimen and was no factor in the sterility.

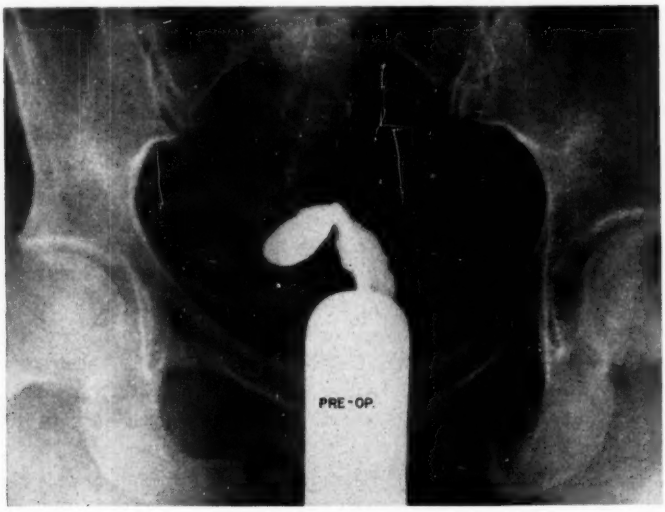

Fig. 1.—Preoperative hysterosalpingogram.

The patient reported that an unplanned conception had occurred the first month after marriage. This was terminated by an induced abortion in 1944 at six weeks' gestation. This was followed by a postabortal, severe pelvic infection and right salpingitis, which were treated conservatively with antibiotics, sulfonamides, and pelvic diathermy. Two years later, in 1946, a second planned pregnancy occurred, after two months' efforts. However, implantation occurred in the right tube, with tubal rupture at six weeks and severe hemorrhage, necessitating surgery. Report of this operation for ectopic pregnancy, which was done elsewhere, stated that the right tube and ovary were completely excised and that transfusions were given. The patient was extremely anxious for a pregnancy, and, after 18 months' failure to conceive following her ectopic operation, she finally sought medical aid for her secondary sterility.

In June, 1948, a Rubin test and a hysterosalpingogram revealed tubal obstruction and no visualization of the remaining left tube. Repeated insufflations done periodically until 1950 showed closed tube. Conservative medical treatment during these two years included thyroid therapy, pelvic diathermy, injections of foreign protein, several series of injections of Jacobson's solution (a solution of ethyl cinnamate in benzyl alcohol and olive oil), vitamins, and psycho-

analysis. The discouraged patient requested pelvic surgery as a final measure with the hope of having tubal patency restored and of possibly becoming pregnant.

The patient's physical examination revealed normal body build, normal breast development and female hair distribution. Weight was 131 lb. (59 kg.), height 61½ in. (156 cm.). The rest of the results of examination were normal. Pelvic examination disclosed a marital introitus, a clean small nulliparous cervix, freely movable, with a small stenotic os. The uterus was normal in size, anterior in position and drawn to the right. There were no adnexal masses felt. There was a midline laparotomy scar of the ectopic surgery, and also a right rectus scar of a previous appendectomy. Laboratory findings reported were: basal metabolic rate, —31%; result of Mazzini test, negative; blood type B, Rh positive; blood count and sedimentation rate, normal; urine, normal. Menstruation had begun at 11 years of age, was regular every 28 days, and lasted 4 days, with severe dysmenorrhea since the menarche. Basal temperature records showed biphasic presumptive evidence of ovulation. Marital relations were reported as normal and had utilized the fertile period. A positive reaction to the Hühner test was reported.

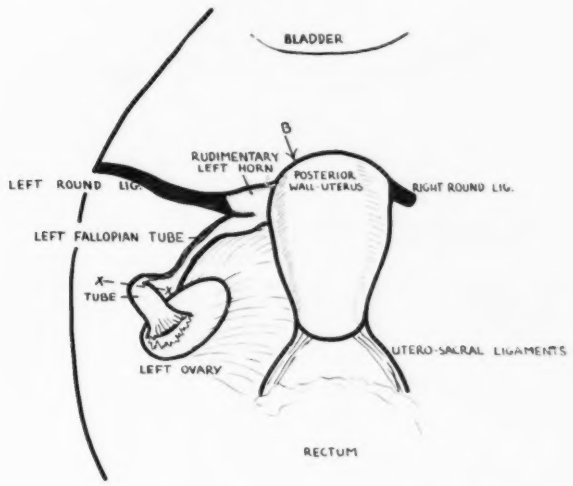

Fig. 2.—Condition found on laparotomy, May 24, 1950.

Operative Procedure and Technique.—On May 24, 1950, at Jewish Memorial Hospital, we performed a laparotomy, with the patient under general anesthesia. Before the laparotomy, the cervix was dilated, and a dressing forceps was inserted into the uterine cavity and held in place by tight vaginal packing. This was in preparation for grasping and withdrawing plastic tubing through the cervix into the vagina after its implantation in the laparotomy opening to be described. The patient was then draped for abdominal operation. The previous midline scar was excised. When the fascia was opened, wire sutures of the previous operation were encountered and were removed. The peritoneum was opened. A loop of small bowel was found loosely adherent near the peritoneum and was freed. The sigmoid was then freed from the body of the uterus and its raw wall was peritonealized with atraumatic sutures. The pelvic pathology was then visualized as follows: The right tube and ovary were missing from previous salpingo-oophorectomy necessitated by ectopic gestation. The fundus of the uterus leaned to the right and there was a rudimentary horn about 1 in. (2.5 cm.) long and ½ in. (1.3 cm.) thick coming off the left side of a didelphic uterus. There was a congenital malposition of the left round ligament, the mesial end of which was confluent with the left uterine horn. The fimbriated end of the tube was patent, and the ampulla was sacculated. The tube was tested for patency by injecting some sterile saline solution into the fimbriated end, and was found to be open for about 1 in. The rest of the tube was atretic. The left ovary appeared normal (Fig. 2).

A long piece of polyethylene (Dupont plastic) tubing, No. 4, which had been previously sterilized overnight in benzalkonium (Zephiran) chloride solution, was inserted as a catheter through the fimbriated end of the left Fallopian tube into the ampulla as far as it could go. The ampulla was then opened over this catheter at the site of the obstruction, and the catheter was pulled through. A Sovak reamer was then used to bore a hole in the left cornual area of the uterus just above and behind the left rudimentary didelphic horn.[2] The piece of tissue that was reamed out, containing uterine wall and endometrial tissue, was sent to the laboratory for examination. The cut end of the resected patent segment of Fallopian tube with the intubated plastic tubing was then inserted part way into the artificial uterine opening, with the polyethylene tubing extending into the endometrial cavity and out through the cervix (Fig. 3).

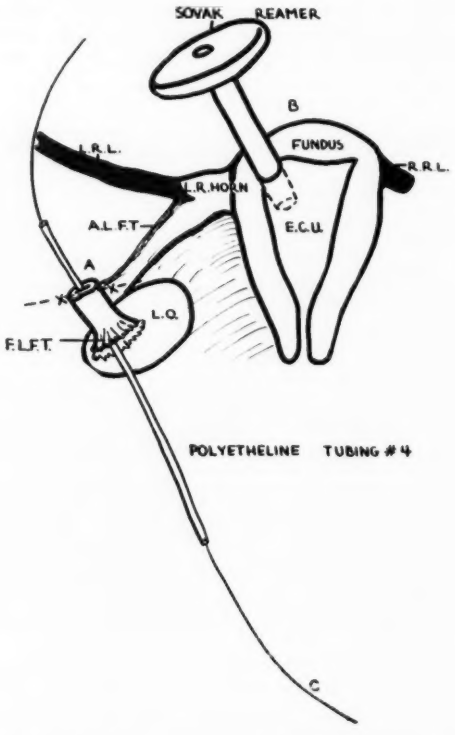

Fig. 3.—Operative procedure, May 24, 1950. Cut end of resected patent segment of Fallopian tube with intubated plastic tubing inserted into artificial uterine opening, with polyethylene tubing extending out through cervix.

It was grasped there by the previously placed dressing forceps and drawn outside the vagina. The tubal stump was held in position in the artificial uterine stoma by two 00 chromic absorbable surgical sutures taken through the tubal serosa and uterine wall. The peritoneal surface of the uterus was closed around the tube with three interrupted sutures, which controlled all bleeding from this site. A redundant loop of atretic tube and broad ligament was formed by bringing the short section of patulous tube and ovary so close to the uterus. This loop was closed over by four interrupted sutures in order to prevent any internal herniation of bowel in this area.

2. Swendson, J. J.: Intrauterine Implantation of Oviducts for Cornual Occlusion by a Reamer Technique, with Report of 4 Cases, Am. J. Obst. & Gynec. **65**:150-157 (Jan.) 1953.

The abdomen was closed in layers, care being taken to bring the polyethylene tube outside the abdominal wall through the lower angle of the operative incision. Consequently the polyethylene catheter extended through the abdominal incision into the fimbriated patent, shortened segment of tube which had been partially implanted into the uterine cornua, emerging through

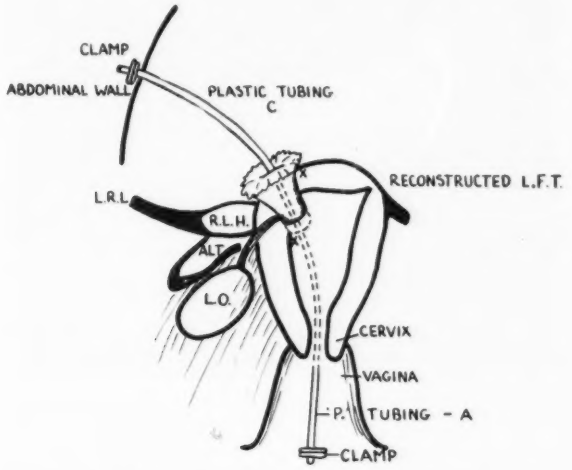

Fig. 4.—Complete tuboplasty, with plastic tube in place.

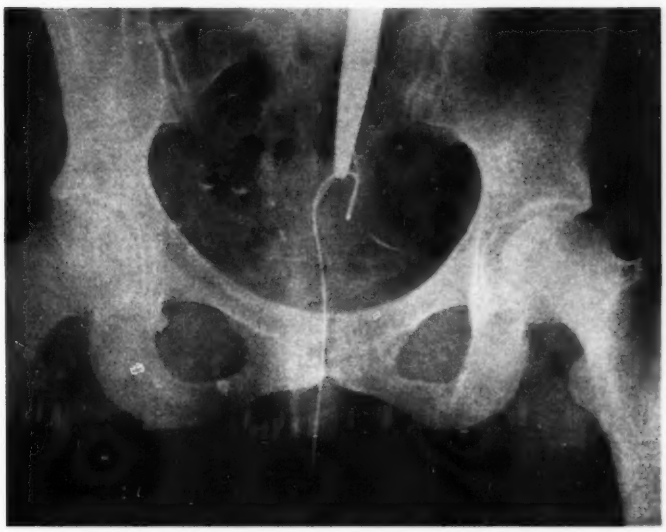

Fig. 5.—Roentgenogram showing intact plastic catheter, anterior view.

the uterine cavity and cervical canal into the vagina (Fig. 4). The plastic tubing was then clamped at both ends and moved freely. The tubing was moved back and forth daily to prevent kinking and adhesions until its removal. On the ninth postoperative day, a flat plate of the abdomen was taken which showed the plastic tubing to be nonopaque. Lipiodol was then

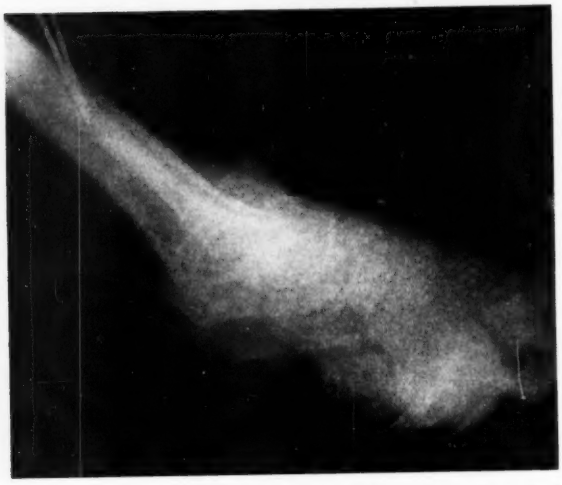

Fig. 6.—Roentgenogram showing intact plastic catheter, lateral view.

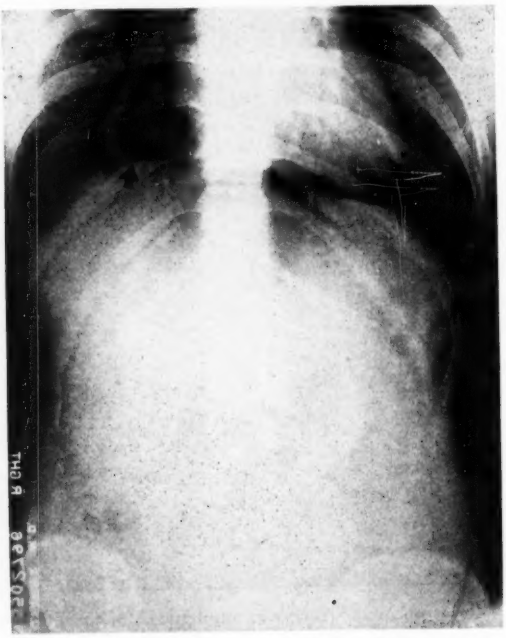

Fig. 7.—Roentgenogram showing pneumoperitoneum.

injected into the polyethylene tubing and both ends were clamped. X-ray photographs taken after this outlined the entire extent of the intact plastic catheter (Figs. 5 and 6). The patient received daily penicillin injections and was out of bed 24 hours after operation. She was discharged on the 10th postoperative day, having made an uneventful, nonmorbid recovery.

The last menstrual period prior to the operation was on April 30, 1950. The menstrual period following surgery began on June 9, 1950, 17 days postoperatively. The polyethylene tubing was removed on June 14, 1950, 21 days postoperatively. Two days after removal of the plastic tubing, a Rubin test was done on the eighth day of the menstrual cycle. This was on June 16, 1950, 23 days postoperatively. The first insufflation showed a closed tube. After administration of β-diethylaminoethyl fluorene-9-carboxylate hydrochloride (Pavatrine) and phenobarbital, and dilation of a slight endocervical stenosis, insufflation revealed tubal patency. This was diagnosed by auscultation and by visualization of a pneumoperitoneum by fluoroscopy and also by roentgenography, with the patient in an upright position (Fig. 7). Kymographic tracing showed patency with CO_2 gas passing at 60 mm. with good tubal contractions (Fig. 8). The patient experienced strong bilateral shoulder pains for the first time. She then had to return to her home in another state. She was advised to resume marital relations on the 10th day of her menstrual cycle, which was 24 days after operation. She was

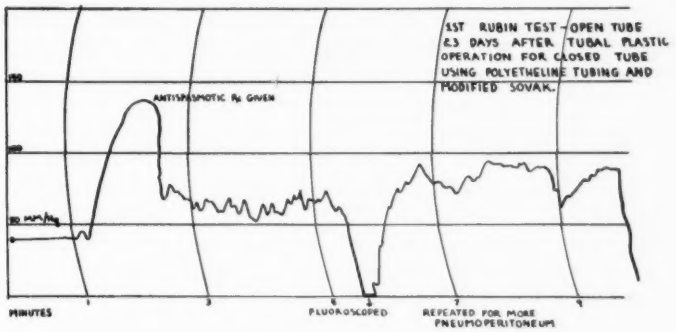

Fig. 8.—Kymographic tracing, showing good tubal contractions.

instructed to have monthly tubal insufflations following each menstrual period to maintain tubal patency if pregnancy did not occur. Despite the fact that temporary tubal patency had been established, she unfortunately failed to conceive that month. Rubin tests done the following month and subsequently have disclosed no further patency. She returned to New York in October, 1950, after further diathermy and Jacobson's solution injections for several months, and numerous subsequent insufflations[3] and x-rays in October, 1950, January, 1951, and July, 1951, revealed no further patency. Follow-up 21 months later disclosed subsequent secondary obstruction of the reconstructed tube, which had definitely been temporarily patent after surgery and immediately after removal of the plastic catheter.

At this time the patient is still willing to undergo further surgery if she could feel assured of a good possibility of repeated restoration of tubal patency. In view of her three laparotomies with postoperative adhesions, further attempts have been deemed inadvisable. She was advised to adopt a child and has applied to an agency.

The biopsy of the reamed-out uterine tissue showed endometrium in follicular phase with hypertrophic muscle fibers.

3. Rubin, I. C.: Uterotubal Insufflation: A Clinical Diagnostic Method of Determining the Tubal Factor in Sterility, Including Therapeutic Aspects and Comparative Notes on Hysterosalpinography, St. Louis, C. V. Mosby Company, 1947.

COMMENT

Original animal experiments proved that plastic tubing used as catheters afforded the best possibility for maintaining patency during recanalization and regeneration of closed Fallopian tubes and for the creation of a permanent lumen in an artificial uterine stoma. Polyethylene tubing was found to be an inert material causing no untoward tissue reaction.[4] Of all substances used as catheters, the polyethylene tubing gave the most normal-appearing regenerated serosa with patent lumen and normal mucosa of the endosalpinx.

No evidence of any harmful body reactions in the use of polyethylene catheters in human pelvic surgery has been reported.[5] A considerable number of successful reconstructed tubal patencies have been reported, with a widely varying percentage resulting in successful pregnancies.[6]

The literature concerning the use of polyethylene tubing discloses a lack of detailed technical information, rather inadequate follow-up studies, and no established uniform criteria for comparative and scientific evaluation of the end-results. The published results on humans show a relatively fair percentage of reestablished tubal patencies and a disappointingly low percentage of pregnancies. However, even the limited number of successes warrants further efforts for the development of improved methods and techniques with this new material. A vital factor which has not yet been satisfactorily determined is the optimum length of time the polyethylene tubing should remain in place in order to assure adequate canalization of a new lumen that will remain permanently patent, and also what measures might prevent secondary tubal closure.[7] The plastic tubing, according to most reports, has been allowed to remain in place approximately two weeks. The best time to perform tubal patency tests postoperatively is still undetermined.[1e] Castallo's work on monkeys showing normal tubal histology when the plastic tubing was allowed to remain in place for one month would seem to indicate that it might be sound in human surgery to allow the plastic tubing to remain in situ for at least one month to ensure firmer serosal and mucosal regeneration that would not fibrose with later secondary obstruction. D'Ingianni reported allowing an intubating cannula to remain in the uterine cornua for three to four months.[8]

4. (*a*) Hellman, L. M.: Use of Polyethylene in Human Plastic Operations, Fertility & Sterility **2**:498-504 (Dec.) 1951. (*b*) Castallo, M.: Recanalization of Fallopian Tubes in Monkey, Fertility & Sterility **1**:435-442 (Sept.) 1950. (*c*) Perl, J.: Polyethylene Intubation in Reimplantation of the Oviducts, J. Internat. Coll. Surgeons **15**:576-584 (May) 1951. (*d*) D'Ingianni, V.: Reconstructive Surgery of Fallopian Tubes Employing a Cannula, South. M. J. **41**:575-581 (July) 1948. (*e*) Wexler, D. J.; Birnberg, C. H., and Kohn, A.: Salpingoplasty Using Polyethylene Tubing: Preliminary Report, Am. J. Surg. **82**:280-282 (Aug.) 1951.

5. (*a*) Hellman.[4a] (*b*) Perl.[4c] (*c*) Greenhill, J. P., Editor: The 1950 Year Book of Obstetrics and Gynecology, Chicago, Year Book Publishers, Inc., 1950, Editor's Note, p. 357.

6. (*a*) Footnotes 1,[a-g] 4,[a,c] and 5.[c] (*b*) Greenhill, J. P., Editor: The 1948 Year Book of Obstetrics and Gynecology, Chicago, Year Book Publishers, Inc., 1949, Editor's Note, p. 341. (*c*) Weisman, A.: Rapidity of Human Fallopian Tube Wound Healing as a Cause for Poor Results from Reparative Tubal Surgery, Am. J. Surg. **82**:278-279 (Aug.) 1951. (*d*) Wexler.[4e] (*e*) Holden.[1h] (*f*) Te Linde, R. W.: Operative Gynecology, Philadelphia, J. B. Lippincott Company, 1953, pp. 616-617.

7. Goldberger, M. A.: Gynecologic Surgery, London, Oxford University Press, 1942, pp. 141-143. Hellman.[4a] Castallo.[4b] D'Ingianni.[4d] Weisman.[6c]

8. Meigs.[1f] D'Ingianni.[4d]

CONCLUSIONS

Tubal patency was definitely reestablished in this case after reparative tubouterine surgery, using polyethylene tubing as a temporary catheter. The plastic tubing was freely movable in the artificial uterine stoma and reconstructed implanted tube for three weeks postoperatively. The tubing was not adherent, and was easily and painlessly removed in the office via the vaginal end, after the outer end was cut flush with the abdominal wall. The removed plastic tubing was intact and smooth and had undergone no change, after being intraperitoneal for three weeks.

In retrospect, although a pregnancy did not result, we, as well as the patient, felt some satisfaction in the reestablishment of tubal patency, even though it was only temporary. It is felt now that the plastic tubing was removed too soon, before good permanent fistulization of the uterine ostium was established. One might take a lesson from reconstructive surgery of the common bile duct, in which the catheters are left in situ for as long as a year. In this case, since we were pioneering, there was some anxiety about the possibility of intestinal obstruction around the tubing and the possibility of infection or irritation while the tubing was in situ. To avert this, the patient was given chemotherapy and antibiotics throughout this period.

The results in this case indicate that polyethylene tubing is an inert material and may be successfully used as a nonadherent catheter in tubouterine plastic surgery for the purpose of attempting reestablishment of tubal patency.

BOWEL PERFORATION FOLLOWING ENEMA THROUGH A PERMANENT COLOSTOMY

PAUL A. KAUFMAN, M.D.
AND
HYMAN SWERDLOW, M.D.
LOS ANGELES

RUPTURE of the intestine has interested military and civilian surgeons since antiquity and has been described as due to a wide variety of causes ranging from penetrating abdominal wounds to comparatively nonviolent muscular effort.[1] Review of the literature would indicate a paucity of reports on bowel rupture following colon irrigations. In 1952 Greene and Greene [2] reported an instance of bowel perforation during the course of a colostomy irrigation, stating that they were able to find only one prior reference [3] to this condition in the literature. Shortly thereafter, Berk [4] described another instance, and more recently Bettman, Richter, and Drugas [5] have added another case report. We believe, however, that there must exist numerous unreported episodes of this kind, since Gabriel [6] was able to collect eight such accidents in the experience at the Saint Mark's Hospital of London in the period between 1937 and 1944, and we have encountered two similar cases in a single year. Zheutlin, Lasser, and Rigler [7] in a recent poll of 100 teaching centers in this country were able to receive reports of 12 perforations in the course of barium enemas via colostomy.

REPORT OF CASES

CASE 1.—M. Y., a 61-year-old white man, had had an abdominoperineal resection for carcinoma of the rectum in 1944. He then remained well, controlling his permanent left iliac colostomy by daily irrigations, although he experienced occasional difficulty in threading the enema catheter into the terminal sigmoid loop. On the morning of Sept. 19, 1951, he was unable to obtain a return of the 1½ to 2 qt. (1.4 to 1.9 liters) of warm tap water with which he had performed his usual gravity irrigation. He started a second irrigation, using additional pressure by means of a rubber hand bulb, and had inserted approximately 100 cc. of water when he experienced sudden pain in the lower abdomen with radiation to the penis and left testis. There was no return of the irrigation fluid through the colostomy.

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3. Davis, C., Jr.: Enema Tube Perforation of the Colon, Ann. Surg. **126**:377, 1947.

4. Berk, J. E.: Perforation Following Barium Enema, Correspondence, J. A. M. A. **148**: 766, 1952.

5. Bettman, R. B.; Richter, H. M., Jr., and Drugas, T.: Perforation of Colostomy Loop by Soft Rubber Catheter, J. A. M. A. **151**:206, 1953.

6. Gabriel, W. B.: Discussion on Management of the Permanent Colostomy, Proc. Roy. Soc. Med. **38**:692, 1945.

7. Zheutlin, N.; Lasser, E. C., and Rigler, L. G.: Clinical Studies on Effect of Barium in the Peritoneal Cavity Following Rupture of the Colon, Surgery **32**:967, 1952.

He was seen about one hour after the incident at which time he presented marked tenderness and rigidity throughout the entire abdomen, more markedly so in the left lower quadrant. The temperature was 99.2 F.; pulse rate, 100, and blood pressure, 154/88. A diagnosis of traumatic rupture of the colon was made, and immediate hospitalization was effected. By the time he was admitted to the hospital the pain had subsided considerably and the initial severe signs of peritoneal irritation had abated, with diminution of tenderness and rigidity. There remained some residual soreness in both lower quadrants, however, and later chills, nausea, and vomiting developed. The white blood cell count on admission was 9,300, with 69% neutrophiles (15% immature forms). Several hours later the white blood cell count had risen to 14,400, with 63% neutrophiles. X-rays of the abdomen at this time revealed no free air under the diaphragm. Examination of the urine revealed nothing abnormal.

Laparotomy was performed on Sept. 19, 1951, at which time a 1 cm. perforation was encountered in the wall of the colon just proximal to its exit from the peritoneal cavity, with gross peritoneal contamination. The area of perforation exhibited no evidence of preexisting disease and was clearly due to trauma. It was sutured and the terminal portion of the sigmoid colon was mobilized sufficiently to permit the sutured area to lie in the extraperitoneal part of the abdominal wound.

On Sept. 23, a partial evisceration occurred around the colostomy site and a second laparotomy was performed for its correction. At the same time a loop of transverse colon was exteriorized, but not opened, to provide for future defunctionalization of the distal colon in the event that this became necessary. Thereafter the postoperative course was quite favorable, and it subsequently developed that the transverse colostomy was never required, the unopened loop of transverse colon being replaced into the abdomen one month later. Further convalescence was uneventful.

Case 2. F. C., a 68-year-old white man, had had an abdominoperineal resection for carcinoma of the rectum on May 15, 1952. He also had severe arteriosclerotic heart disease with auricular fibrillation. On July 11, a long pedunculated polyp was observed at the colostomy stoma, and this was excised completely. Microscopic examination revealed early adenocarcinoma restricted to the tip of the polyp. Further colonic work-up was interrupted because of urgent urologic obstructive symptoms, for which transurethral prostatic resection was advised. On admission to the Cedars of Lebanon Hospital, however, he was found to be in cardiac decompensation with uncontrolled auricular fibrillation. Preoperative preparation including full digitalization did not control these conditions adequately until Aug. 5, when his status improved to the extent that a transurethral resection could be done with relative safety.

On Aug. 8, a barium enema was done. A bag catheter was used to administer the enema through the colostomy to prevent reflux of the barium suspension. During the course of the fluoroscopic examination the patient complained of severe abdominal pain. X-rays taken revealed barium in the free abdominal cavity.

Various consultants were at odds regarding the immediate management of the patient. It was finally decided that, primarily because of his cardiac status, it would be best to institute conservative measures, with close observation. These consisted of nasogastric suction, parenteral feeding, transfusions, antibiotics heavily administered by all routes, digitalis therapy, and analgesia.

Symptoms gradually subsided under this regimen, with progressive clinical improvement each day. The highest temperature was 101 F.; otherwise, he was essentially afebrile. The colostomy seemed to function fairly well, feces being passed readily. However, much barium remained in the ascending and descending colon. By Aug. 18, he had seemingly responded well enough to be given a soft diet and to be ambulating gradually.

On Aug. 20, just 12 days after the perforation, he suddenly complained of severe lower abdominal pain, and signs of peritonitis and shock rapidly developed, precluding any attempt at surgical intervention. Death ensued two days later.

Postmortem examination revealed widespread fibrous adhesions throughout the peritoneal cavity, which kinked, but did not obstruct, the small intestine in many places. There were about 50 cc of purulent exudate and barium in the left lower quadrant and over the colon in the region of the stoma. The colon was so friable and necrotic for a distance of 8 cm. adjoining the stoma

that no definite perforation site could be made out. There was a 5 mm. thin pedunculated polypoid nodule 5 cm. proximal to the stoma. There were numerous minute diverticula in the 20 cm. of colon proximal to the stoma.

COMMENT

No data are available in any of the published reports as to the actual hydrostatic pressure attained by the enema at the time of perforation, and, although considerable force was used in one of our cases, most of the other reports would indicate that no undue pressure had been applied. A somewhat more significant observation emerges from this study, however, and that is the constant location of the rupture at the bend of the terminal loop of bowel a short distance proximal to the colostomy. It is at this point that the direction of the sigmoid changes sharply as it bends forward at a right angle to make its exit from the abdomen.

It is a common clinical observation that this tortuosity in the course of the terminal loop of colon occasionally causes some difficulty during insertion of the enema tube,[5] and patients frequently report that it may require considerable manipulation before the catheter can be inserted into the colon a sufficient distance to prevent reflux of the irrigating fluid. It is entirely conceivable therefore that an inept insertion of the enema tube may damage or actually perforate the colon at the point of its greatest angulation and that further manipulation may then effect the passage of the enema tube into the more proximal colon without the patient's awareness of the damage that had been done. According to this reconstruction of the sequence of events, the mechanical laceration of the bowel wall may not be signaled by any unusual pain, since intestine is insensitive to this kind of stimulus, but the gross contamination of the peritoneal cavity then occurs as a result of the hydrostatic pressure built up by the instilled fluid and its efflux through the perforation.

The actual peritoneal soiling is often heralded by the sensation that "something has given way" in the abdomen, with immediate severe abdominal pain and mild shock. There frequently ensues a deceiving improvement in the patient's general condition for several hours followed by the reappearance of signs of peritoneal irritation and a constitutional reaction presaging severe shock.

Treatment should be instituted at the earliest possible moment, since fecal contamination is often very severe and continues as long as the perforation can serve as a feeding focus. Davis [3] treated the bowel laceration by intraperitoneal suture in two patients upon whom he operated immediately, with good results. His third patient was successfully treated nonoperatively, but a review of the published protocol casts considerable doubt on the diagnosis of bowel perforation which was never established unequivocably. In the cases reported by Greene and Greene,[2] Berk,[4] and Bettman [5] and his associates, it was possible to mobilize the colon sufficiently to permit resection of the distal portion of the colon, including the perforated area, with reestablishment of a new colostomy. In our first case it was not possible to mobilize sufficient colon to accomplish this readily, although suture of the perforation was effected and the damaged area of the bowel was brought outside the parietal peritoneum. In each of these cases recovery ensued. Gabriel [6] reports on two patients similarly treated, with fatal results, but it is believed that none of these patients was treated early. On the other hand, three of his patients and our second patient, who were not operated upon, all died, as did three of his patients in whom a proximal defunctionalizing colostomy was performed without closure of the perforation. It therefore appears that exteriorization or closure of the perforation is the

sine qua non of any program of treatment of perforation of the colon, whether it is accompanied by proximal defunctionalizing colostomy or not. Antibiotic therapy, transfusions, electrolyte and fluid administration, and other supporting measures are also indicated. In our opinion, the urgency of operative treatment in this situation is such as to override other considerations of the patient's general condition, even if, as in our second case, the cardiac status causes concern over the patient's ability to withstand major surgery. We believe that patients who represent poor operative risks are even less able to cope with unrelieved peritoneal soiling than with the trauma of operative intervention. Antibiotic therapy without surgery may mask severe peritonitis temporarily with disastrous late consequences. Early diagnosis and immediate exteriorization of the damaged area, or its secure closure, with or without a defunctionalizing colostomy represent the most effective management of enema-induced colon rupture in the light of our present experience.

SUMMARY

Two cases of colon rupture following enema through a colostomy are reported, and the literature on this subject is reviewed. It is postulated that perforation at the bend of the sigmoid immediately proximal to the colostomy is produced mechanically by the tip of the enema tube and that peritoneal contamination is then effected when the irrigating fluid is introduced. Treatment recommended is immediate laparotomy and exteriorization of the perforated bowel by establishment of a new colostomy at the level of the perforation.

REPEATED SUCCESSFUL FEMORAL EMBOLECTOMY

Report of a Case

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AND
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OCCLUSION of a major peripheral artery by an embolus is a serious emergency all too often leading to gangrene of the involved extremity and resulting in permanent disability or in death of the patient.

Until 1911, when Labey [1] performed the first successful embolectomy, little had been accomplished toward reducing the loss of life and limb associated with arterial embolization. Since that time many excellent reports of collected series of embolectomies have been published.

Reports of repeated successful removals of emboli from the same vessel in one patient are of sufficient rarity and interest to warrant comment. It is the purpose of this paper to present such a case with the hope that it may stimulate interest in direct surgical attack upon recurrent emboli even in the very poor-risk patient.

REPORT OF A CASE

P. H., a 66-year-old white man, was admitted to the surgical service of the King County Hospital on Jan. 2, 1952, six hours after the onset of sudden sharp pain in the left foot and lower leg, followed by coldness, numbness, and reddish-purple discoloration to the level of the knee. He had been under treatment for valvular heart disease for the previous five years and had severe dyspnea and orthopnea for five days prior to admission.

Examination revealed auricular fibrillation with moderate cardiac decompensation. The left leg was cooler than the right and was of a reddish-purple hue to the level of the knee. A very weak femoral pulse was palpable at the level of the inguinal ligament, but no pulses were palpable below this. Good femoral, popliteal, and dorsalis pedis pulses were palpable on the right. A diagnosis of embolic occlusion of the left femoral artery was made. Left lumbar sympathetic block was immediately carried out, with no change in temperature or in the appearance of the lower leg.

First Operation (Jan. 2, 1952).—Four hours after admission to the hospital and 10 hours after the onset of symptoms, embolectomy was performed by one of us (H. N. H.) with the patient under local anesthesia. The left femoral artery was exposed in the femoral triangle. No pulsations were present. A transverse arteriotomy was made, and by gentle suction a clot was evacuated both proximally and distally. A free flow of blood, both proximally and distally, followed this procedure. The arterial incision was closed with a continuous over-and-over suture of 5-0 nonabsorbable surgical (silk) suture material. The left popliteal, but not the dorsalis pedis, pulse was palpable after the operation.

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Fig. 1.—Emboli removed from left femoral artery at time of second embolectomy.

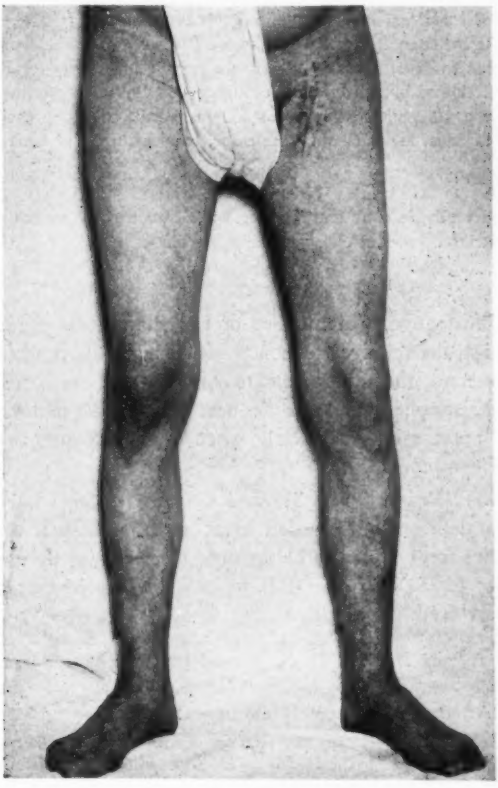

Fig. 2.—Appearance of extremities 14 days after second embolectomy.

Postoperatively, heparin was given in adequate dosage to maintain the clotting time at 15 minutes until bishydroxycoumarin (Dicumarol) therapy became effective. Bishydroxycoumarin therapy was continued throughout the next month. Convalescence was marred by the development of a pulmonary embolus on the second postoperative day and a stitch abscess on the seventh day. Auricular appendectomy was considered, but this procedure was rejected by the patient. He was discharged on the 51st hospital day. Femoral and popliteal pulses were palpable at this time.

In May, 1952, he complained of pain in the left leg on walking more than 500 ft. (152.4 meters). Lumbar sympathetic block caused subjective improvement and increase in skin temperature. Lumbar sympathectomy was offered and refused.

The next hospital admission was on Oct. 23, 1952, because of severe cardiac decompensation. At 1:00 p. m. on the following day he experienced sudden excruciating pain in the left foot and leg, with rapid development of numbness, coldness, and a bluish discoloration to the midthigh. No pulses were palpable on the left lower extremity.

Second Operation (Oct. 24, 1952).—Embolectomy was undertaken by one of us (E. J. S.) two hours after onset of symptoms, with the patient under local anesthesia and with use of the same approach as previously. The femoral artery was found to be in excellent condition with only slight thickness at the site of the previous arteriotomy. A vertical arteriotomy was made, and an embolus consisting of old organized clot (Fig. 1) was removed from the proximal segment, utilizing the catheter "piston" technique of Andrews and Harkins[2] (1932). This was followed by a copious gush of blood. A fresh clot was removed from the vessel below the incision, which was followed by retrograde bleeding. After closure of the incisions, femoral and popliteal pulses were again palpable. Heparin and bishydroxycoumarin therapy was instituted and maintained for 14 days, at which time the color, temperature, and function of the leg were good (Fig. 2).

When the patient was last seen on June 13, 1953, femoral pulses were of good quality, left popliteal weak, and dorsalis pedis absent. The leg was of good color and warm. He complained of intermittent claudication brought on by walking one block.

COMMENT

This case is illustrative of a number of important facts regarding peripheral arterial emboli: first, the relative frequency with which this condition occurs in the cardiac patient; second, the multiplicity of embolic phenomena in many of these cases, and third, the simplicity and effectiveness of operative removal of the embolus with use of local anesthesia, particularly when this procedure is performed early after the onset of symptoms.

Danzis[3] (1933), Pearse[4] (1933), and Garvin[5] (1944) all emphasized the importance of mural thrombi in the left heart in the causation of arterial emboli. The femoral arteries were shown to be the ones most frequently obstructed by such emboli by Danzis[3] (1933). Key[6] (1921 to 1922) indicated that the left side was the side oftenest involved.

2. Andrews, E. A., and Harkins, H. N.: Embolectomy: Report of a Case Involving the Femoral Artery, Ann. Surg. **96**:40-43 (July) 1932.

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6. Key, E.: Uber Embolectomi als Behandlungsmethode bei embolischen Zirculationsstörungen der Extremitäten. Acta Chir. Scandinav. **54**:339-416, 1921-1922.

Multiple embolizations have been reported by Harkins [7] (1934), Deitch [8] (1936), McFarlaine (1940), Reynolds and Jirka [9] (1944), Scott and Williams [10] (1949), McGarity and Robertson [11] (1953), and others.

Most authors writing on the subject of embolectomy agree that the probability of a successful outcome is decidedly less with the lapse of 10 hours or longer after onset of symptoms. Recently, however, Olwin, Dye, and Julian [12] have reported success after intervals as long as three and one-half months. Anticoagulant drugs are of great value in extending the time limit of successful operability as well as increasing the effectiveness of early operation. Pearse [4] (1933) and Linton [13] (1941) emphasized the need for early embolectomy and pointed out that formation of widespread secondary thrombi distal to the embolus render late embolectomy much more prone to failure. Reduced blood flow in the decompensated cardiac patient limits the effectiveness of collaterals, as stressed by Andrus [14] (1950), and also tends to facilitate further blockage by distal thrombus formation.

The frequently associated cardiac disease, age, and often generally poor condition of these patients make local anesthesia the agent of choice. Gentleness and the use of a well-oiled soft rubber suction catheter will dislodge proximal and distal emboli and associated thrombi with a minimum of intimal damage. The use of anticoagulants postoperatively reduces the incidence of thrombosis at the site of arteriotomy as well as the propagation of thrombi which may remain in small distal branches. It would seem wise to continue the use of anticoagulants indefinitely as suggested by Lord and Burke [15] (1953) to help prevent formation of further mural thrombi and subsequent embolization. In selected cases interval mitral commissurotomy and auricular appendectomy would seem indicated. The proper treatment of patients with recurrent peripheral arterial embolization requires the cooperation of general practitioner, cardiologist, and surgeon.

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SPONTANEOUS CHOLECYSTECTOMY

Report of a Case Probably Resulting from Irradiation of Hepatic Metastasis

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THERE are a number of reports in the literature describing spontaneous extrusion of portions of various viscera either via the natural excretory route of the alimentary canal, namely, per rectum, or through operative or traumatic wounds in the body parietes. The prerequisite for passage of portions or all of a viscus spontaneously is the devitalization and sequestration of the viscus. Perhaps the most frequent of these is the passage of a segment of bowel per rectum (sometimes as a "cast") following a volvulus or intussusception. In the following instance practically the entire gangrenous gall bladder without stones was extruded spontaneously through the abdominal wound following left hepatic lobectomy, performed several weeks previously for the excision of hepatic metastases, which was performed in turn after irradiation of the left lobe and porta hepatis.

REPORT OF CASE

A. G., a man aged 67 years, in 1943 underwent a radical gastrectomy for carcinoma of the midportion of the stomach in another institution. His clinical course was satisfactory until the spring of 1952, at which time a second laparotomy was performed because of upper abdominal pain and vomiting. On this occasion a resection of an upper segment of jejunum was purported to have been carried out because of a metastasis in its mesentery causing occlusion of the lumen. A "nodule" was noted in the left lobe of the liver, but nothing was done about this. During the summer and early fall of 1952 some relief from upper abdominal pain was experienced, but this again became pronounced, and finally a large mass was palpable in the region of the left lobe of the liver. It was assumed that this was a metastasis, and in a third institution irradiation therapy in "larger doses" was given through a square portal centering over the mass and extending across the midline. This therapy was completed by the end of December, 1952. The technical factors were as follows: 8 by 6 cm. portal centered over the mass in the left lobe of the liver; 250 kv.; 50 cm. target-skin distance; half-value layer 2.2 mm Cu.; 2,700 r estimated tumor dose at 6 cm. below the skin; skin dose 5,500 r.

The patient was seen by me on Jan. 15, 1953, at which time a fist-sized mass was palpable in the left upper quadrant. He complained of more or less constant deep upper abdominal pain that had severe exacerbation. On Jan. 27, 1953, laparotomy was performed through a long transverse incision, with vertical component near the midline. The entire left lobe of the liver was resected, because it was almost completely replaced by a metastatic mass. The line of transection was just to the left of the falciform ligament. Five discrete metastases were excised from the right lobe. The gall bladder was noted to be normal and was left in situ. The remainder of the abdomen was free from cancer.

Histologic study of the resected masses showed anaplastic carcinoma. Sections of the carcinoma removed 10 years previously also showed anaplastic carcinoma. (These sections were kindly loaned by the Department of Pathology of the Hospital for Joint Diseases, New York.)

The patient was discharged from the hospital on the 21st day. During the convalescence an intense cutaneous reaction developed due to the previously given radiation. This resulted in a partial dehiscence of the vertical component of the latest laparotomy wound. The limited dehiscence became converted into an indolent ulcer 6 by 4 cm. in size.

From the Memorial Center for Cancer and Allied Diseases.

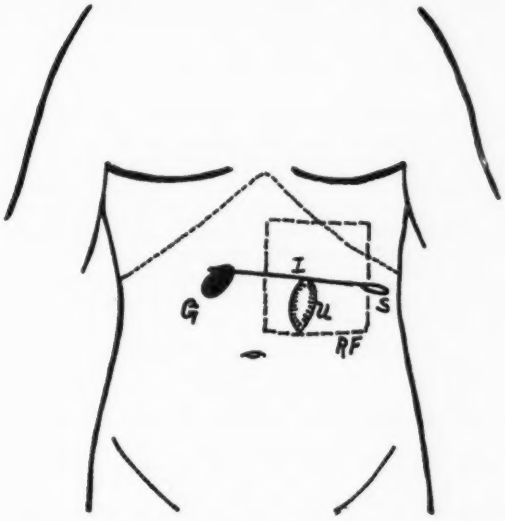

Fig. 1.—Diagram showing abdomen at the time the spontaneously separated gall bladder was found partially extruding from recent laparotomy incision. *I*, transverse incision for left hepatic lobectomy; *RF*, square-shaped radiation therapy portal; *U*, indolent ulcer produced by separation of vertical component of laparotomy incision; *S*, nearly healed sinus from which subdiaphragmatic abscess drained spontaneously; *G*, partially extruded gangrenous gall bladder in sinus that opened in extreme right portion of transverse incision.

Fig. 2.—Photograph of partially opened, spontaneously extruded gall bladder. *C*, stump of cystic duct.

Pain was relieved, and his condition appeared satisfactory until March 1, 1953, when he was readmitted to the hospital because of rise in temperature to 103 F. (oral). The following day green purulent material was discharged from the extreme left angle of the transverse incision; the temperature became normal, and in five days he was sent home. Five days later at an office visit the sinus in the left extremity of the transverse wound appeared to have closed, but a similar sinus discharging bile-stained pus was noted in the right portion of the transverse wound; dressings were changed. Seven days later at a second office visit there was a black piece of tissue partially extruding from the previously noted sinus; this was readily removed and appeared to be a collapsed sequestrated gall bladder (Figs. 1 and 2). Following its removal, clear greenish bile escaped. The gall bladder was cut open, placed in formalin, and sent to the pathology laboratory. In the fresh state it measured 8 by 5 cm. The report received from the laboratory stated: "Specimen consists of a green-stained sac, 6 by 4.5 cm. The surface is granular and there is a small aperture at one end presumably the cystic duct of the gall bladder. The wall of the sac is uniformly 2 mm. in thickness and the lining smooth. 2 sections. Diagnosis: Acute inflammation in a gangrenous sac which is consistent with the gall bladder." A week later the patient was seen, and the sinuses at each extremity of the transverse laparotomy wound were almost completely healed. The chronic ulcer in the irradiated field appeared indolent. On April 24, 1953, the patient was again examined and had gained 18 lb. (8.2 kg.) since operation. He was free from pain and had resumed brief daily visits to his place of business. In September, 1953, his improvement was maintained, and he continues to lead a normal existence.

COMMENT

The series of events that led to the spontaneous gangrene, sequestration, and extrusion of the gall bladder may be summarized as follows: A quantity of radiation was delivered to the region of the porta hepatis. A short time later a certain measure of trauma was sustained by the region incident to the left hepatic lobectomy before the full effects of the irradiation were manifested in the tissues. The latter changes then occurred and included vascular occlusion to the gall bladder. This viscus then became progressively gangrenous, and an abscess developed about it. The abscess drained itself through the recent operative wound, and the then spontaneously sequestrated gall bladder extruded itself through a sinus in the laparotomy wound.

HYGROMA COLLI OF THE NEWBORN INFANT INVOLVING THE FLOOR OF THE MOUTH

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CREDIT is usually given to Redenbacher [1] for having published in 1828 the first description of a cystic hygroma, an uncommon congenital cystic endothelium-lined lymphatic tumor most frequently arising in the neck. The occurrence of a cystic hygroma in infants and children can no longer be considered a rarity, a total of 225 cases having been collected from the world literature by Gross and Goeringer [2] in 1939. Since then there have been sporadic reports on the subject. The purpose of this paper is to report two unusual cases of hygroma colli of the newborn infant which were unique by virtue of their rapid growth and extensive invasion of the tongue and floor of the mouth. The management of these infants posed many problems to the attending pediatrician, surgeon, radiotherapist, and anesthetist because of the serious embarrassment of respiration and swallowing which developed in the first weeks of life. The occurrence of hygroma colli of the newborn infant with involvement of the floor of the mouth is a complication of hygroma which has been described but rarely in the literature.[3]

REPORT OF A CASE

CASE 1.—Baby H. was a boy born to a young mother whose pregnancy was uneventful. The baby was delivered by Dr. Stewart Bauman at the Syracuse General Hospital on Oct. 2, 1951, and weighed 5 lb. (2.3 kg.). Physical examination was not remarkable, except for a cystic swelling of the left side of the neck measuring 5 by 3 cm. and which was situated in the submaxillary region of the anterior triangle (Fig. 1). Slight elevation of the floor of the mouth near the sublingual ducts was noted at this time. The mass did not transilluminate. Laboratory and x-ray studies revealed no abnormalities other than a soft tissue swelling of the left side of the neck. Aspiration of the mass on two occasions yielded frank blood which failed to clot. No recession of the tongue elevation was noted. The infant was discharged on Oct. 12, 1951, to be followed closely at home.

On Oct. 17, 1951, the patient was admitted to Syracuse Memorial Hospital because of an increase in the size of the tumor and serious feeding difficulty caused by further elevation of the tongue. The increasing severity of symptoms made surgical intervention mandatory on the 16th day of life. Operation was performed under endotracheal anesthesia. A multilocular cyst was found in the anterior triangle of the neck. It lay in intimate association with the carotid artery and jugular vein. Several large venous tributaries traversed the cyst. Complete excision of the

From the Departments of Surgery and Pediatrics, State University of New York at Syracuse College of Medicine.

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Fig. 1 (Case 1).—Note the rounded swelling in the anterior cervical region just inferior and anterior to the ear lobule.

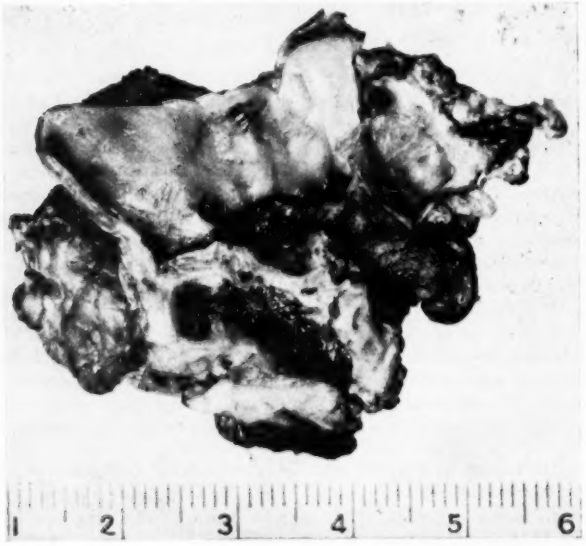

Fig. 2 (Case 1).—Surgical specimen from patient. Note the multiloculated nature of the hygroma and the presence of blood clots within some of the cysts.

cyst was impossible by virtue of a thin stalk of hygromatous tissue which was seen to invade the floor of the mouth. The latter region was swabbed with iodine solution in an effort to sclerose the remaining daughter cysts. At the termination of the operation, which required three hours, the endotracheal tube was withdrawn, and the baby appeared to be free of respiratory distress. It was felt by the consulting otolaryngologist that tracheotomy was not indicated. The usual postoperative supportive measures were instituted. Pathological examination of the surgical specimen revealed it to be a multiloculated cystic hygroma. Several of the larger cysts contained blood clots (Fig. 2). Several hours following the operation the baby became ashen, and severe respiratory distress occurred. Convulsive movements rapidly supervened, and tracheotomy was performed shortly thereafter. The baby's condition, however, did not improve, and he died 12 hours after conclusion of the operation.

The autopsy was performed by Dr. W. D. Alsever, of the Department of Pediatrics, State University of New York at Syracuse, Medical Center. The operative field was dry, and no

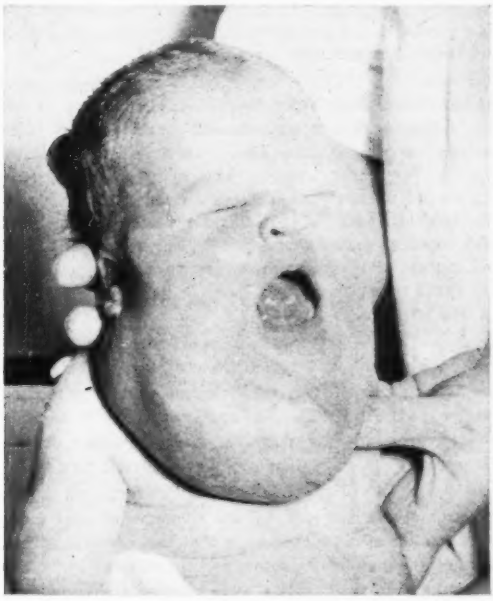

Fig. 3 (Case 2).—Huge swelling of the neck due to hygroma involving anterior cervical triangles. Note the elevation of the tongue toward roof of the mouth.

residual tumor could be recognized in the neck. There was marked edema of the tongue, pharynx, and larynx. There was noted the presence of hygroma in the region of the frenum of the tongue. Microscopic examination revealed invasion of the tongue and floor of the mouth by hygroma. The cause of death was ascribed to acute pulmonary edema and asphyxia.

CASE 2.—Baby S. was delivered by Dr. Stewart Bauman at the Syracuse General Hospital on May 17, 1952. The mother had an uneventful pregnancy, and no difficulty was experienced in the obstetrical management of the case. The baby weighed 9 lb. (4.2 kg.) at birth. Upon delivery of the shoulders, the obstetrician noted a large swelling in the region of the neck. Examination immediately after birth disclosed a huge tumorous enlargement in the anterior cervical region which extended upward bilaterally to involve the submandibular regions. The tongue and floor of the mouth appeared slightly elevated (Fig. 3). There was no interference with respiration or swallowing, and the child, who appeared vigorous, took all feedings during the first few days of life. Chest x-rays and blood studies were within normal limits. On the third day aspiration was performed because of progressive enlargement of the mass and bluish

discoloration of the overlying skin. There was obtained 60 cc. of fluid which appeared frankly bloody. This material did not clot, and laboratory studies revealed merely blood cells and no neoplastic elements. Subsequent aspirations yielded large quantities of similar fluid, as well as clear amber fluid from other sites in the tumor mass. Despite repeated aspirations, the cystic tumor would return to its former size coincidentally with the elevation of the floor of the mouth, most noticeable in the sublingual region. Because of the latter, the child began to have feeding difficulties.

On the fifth day of life radiological consultation was obtained, and a course of radiotherapy was instituted. External radiation, 100 r daily, was delivered to the cervical and submandibular regions (through both lateral and anterior portals) for the next seven days. On the 27th day of life, because of severe respiratory difficulty, a tracheotomy was performed by Dr. A. H. Rubenstein. Temporary alleviation of the respiratory symptoms resulted. Nourishment was provided through a nasogastric polyethylene tube. During the next seven weeks supportive therapy was given, including several blood transfusions, which were required to restore the blood lost through repeated aspirations of the cyst. No beneficial effect of radiotherapy was observed during this period. It was apparent that the tongue and the cervical tumor were rapidly enlarging, and surgical intervention seemed indicated.

On July 9, 1952, (infant was 6 weeks old) excision of the hygromatous mass in the left side of the neck was performed under ether anesthesia administered through the tracheotomy tube. The left anterior cervical triangle was exposed through a long incision extending from the left ear to the midline beneath the chin. There was disclosed a typical giant multiloculated cystic hygroma occupying the anterior triangle. This hygroma extended across the midline to the right cervical region and inferiorly to the suprasternal notch. Several large-sized venous channels connected the hygroma with the jugular vein, to which it was intimately attached. It was virtually impossible to excise the tumor completely, as it extended into the floor of the mouth behind the mylohyoid muscle. Pathological examination of the excised specimen revealed it to be a multiloculated cystic tumor measuring 7 cm. in its greatest diameter. The histological picture was that of cystic hygroma.

Despite the removal of the bulk of the tumor from the left anterior cervical region, there was no recession of the sublingual swelling. On the contrary, the enlargement became greater, and the tongue became so turgid that it protruded noticeably from the mouth. Prolonged periods of apnea occurred in the postoperative period, and convulsions were noted on one occasion. Despite vigorous supportive therapy, the baby had a progressively downhill course and died on Aug. 3, 1952, at the age of 2½ months, four weeks after partial surgical excision of the tumor.

The autopsy was performed by Dr. W. D. Alsever. A large multilocular cystic mass was found in the right anterior cervical region. (There was an incisional scar in the left cervical region.) Straw-colored fluid escaped from some of the cysts, whereas blood was noted in other cysts. The mass infiltrated the floor of the mouth and the tongue. Microscopic sections through the enlarged tongue revealed invasion of its muscle bundles by hygroma.

COMMENT

The differential diagnosis and treatment of cystic hygroma present many problems by reason of its invasive quality. The independent growth potential of this condition has been emphasized by Goetsch,[4] who states that a "hygroma is a benign true neoplastic tumor of lymphatic origin." Most pathologists concur with Goetsch that a hygroma grows by virtue of proliferation of endothelial cells to form cords and that these cords secrete lymph and become vacuolated, the end-result being an invasive cyst. Watson and McCarthy,[5] in pointing out the general high mortality rate of cystic hygroma, regardless of therapy, have stressed that

4. Goetsch, E.: Hygroma Colli Cysticum and Hygroma Axillare: Pathologic and Clinical Study and Report of 12 Cases, Arch. Surg. **36**:394, 1938.

5. Watson, W. L., and McCarthy, W. D.: Blood and Lymph Vessel Tumors: Report of 1,056 Cases, Surg., Gynec. & Obst. **71**:569, 1940.

those hygromata noted at birth seemed to grow at a much faster rate. This observation is pertinent in our cases and should alert the pediatrician to the gravity of this condition in the neonatal period.

The differential diagnosis of tumors situated in the anterior triangle of the neck in the neonatal period should include branchial cleft cyst, thyroglossal duct cyst, hemangioma, teratoma,[6] sarcoma, and submaxillary adenitis. The precise diagnosis often requires surgical excision and pathological examination. Gross and Goeringer, in their series, found that the large majority of cystic hygromata occurred in the posterior triangle of the neck, transilluminated readily, and yielded clear amber fluid on aspiration. It should be noted that our cases were quite atypical, since they satisfied none of these criteria, in that the tumors were situated anteriorly, did not transilluminate, and on aspiration yielded blood which did not clot. Stadler and his associates [7] recently reported in a child a hygroma, also situated in the anterior triangle, which on aspiration produced blood that failed to clot.

In the two cases presented, the management was complicated by two factors: namely, the occurrence of the hygroma at birth and the involvement of the floor of the mouth. While it was recognized that the newborn infant can tolerate major surgery, operation was postponed because of jaundice and weight loss in the first patient. The increased swelling of the tongue and floor of the mouth, rather than the extent of the cystic cervical mass, made surgery mandatory at 16 days of life. Tracheotomy at the completion of surgery was not considered warranted by us in this case because of the baby's apparently good condition. (It is to be remembered that tracheotomy itself in the neonatal period carries a significant mortality rate.) The clinical course and the autopsy findings demonstrated laryngotracheitis incident to intubation. Tracheotomy at the time of surgery might have prevented the fatal outcome in this case. On the basis of his autopsy studies, Eckenhoff [8] has advocated early tracheotomy when difficult respirations first become apparent. Eckenhoff has emphasized the hazards of intubation of the infant larynx, with its consequent laryngeal trauma and edema. He has stressed that the edema of the tiny infant glottis may compromise the airway sufficiently to cause cerebral anoxia in the space of a few hours.

The second case presented many features identical with the first but to a more marked degree. The outward cervical swelling was bilateral and extensive, and the sublingual edema was already evident at birth. The unsuccessful outcome of the first case prompted us to select radiotherapy as an alternative method of management in the second case, despite the generally accepted opinion that a cystic hygroma is relatively radioresistant. Radiotherapy has been reported to be effective in the treatment of cystic hygroma by Hodges and co-workers,[9] Pfahler and Perlman,[10] and

6. McGoon, D. C.: Teratomas of the Neck, S. Clin. North America **32**:1389, 1952.

7. Stadler, H. E.; Berman, J. K., and Johnson, T. W.: Hygroma Colli: A Retropharyngeal Tumor Simulating an Angiomatous Growth, J. Pediat. **41**:100, 1952.

8. Eckenhoff, J. E.: Some Anatomic Considerations of the Infant Larynx Influencing Endotracheal Anesthesia, Anesthesiology **12**:401, 1951.

9. Hodges, F. M.; Snead, L. O., and Berger, R. D.: Roentgen Therapy of Cystic Hygroma of Neck in Children, Am. J. Roentgenol. **42**:551, 1939.

10. Pfahler, G. E., and Perlman, H. H.: Cystic Hygroma of the Neck and Mediastinum Successfully Treated with Roentgen Rays, Am. J. Roentgenol. **63**:539, 1950.

Singleton [11] and has been employed successfully at the Mayo Clinic.[12] The standard prescribed course of 700 r was delivered into the tumor over a period of one week beginning on the fifth day of life. Increasing respiratory and feeding difficulties became profound, necessitating tracheotomy, which was performed on the 27th day of life.

In an attempt to alleviate the distressing elevation of the floor of the mouth and the progressive enlargement of the tongue, surgical excision of the hygromatous mass on the left side of the neck was performed when the infant was 8 weeks old. Both surgery and radiotherapy proved to be completely ineffectual in decompressing the critical sublingual region. The autopsy revealed actual hygromatous invasion of the tongue and floor of the mouth. Under such circumstances excision of the cervical component of the hygroma will not forestall further encroachment in the floor of the mouth. During a 10-week period of observation, radiotherapy also proved ineffectual in this regard. Only the actual excision or destruction of the intraoral component of such a tumor will relieve the critical sublingual swelling. It must be conceded, therefore, that we are dealing with a desperate situation, since such radical surgery in the newborn infant would be mutilating.

CONCLUSION

Two unusual cases of cystic hygroma of the neck occurring in the newborn infant with involvement of the tongue and floor of the mouth have been reported. Problems concerned with the diagnosis and treatment of this condition have been discussed. The presence of sublingual edema in cases of hygroma colli of the newborn infant portends a grave prognosis. Early tracheotomy and operative excision, in stages if necessary, are recommended in the management of such cases. Hygroma colli of the newborn infant with involvement of the tongue and floor of the mouth must be recognized as a condition with a high mortality rate.

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11. Singleton, A. C.: Congenital Lymphatic Diseases—Lymphangiomata, Ann. Surg. **105**:952, 1937.

12. Stark, D. B.: Personal communication to the authors.

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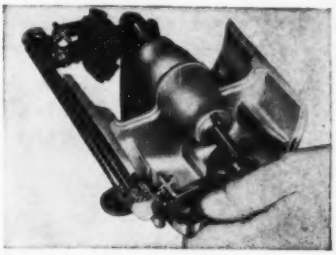

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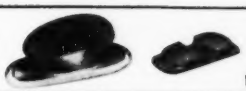

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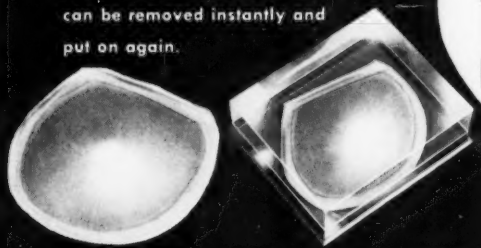

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